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Original Article

Spinal Meningioma: Five-year experience of a single center

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Abstract

Aim: To discuss the natural history of spinal meningiomas (SMs) in our series, surgical approach options, and pathology results in light of the literature.

Material and Methods: Of the 151 meningioma cases operated on between June 2014 and August 2020 in our center, 11 with spinal localization were analyzed retrospectively.

Results: Three (27%) of our cases were male, eight (73%) were female, and 7.2% of all meningioma cases were located in the spine. The patients mostly presented with paraparesis, and 73% of the tumors had a thoracic localization and 64% had a lateral spinal localization. We were able to reach a 90% Simpson grade I resection limit after the opening procedure performed with mostly hemilaminectomy or laminoplasty. The histopathologies of all our cases were reported as meningothelial and transitionally weighted WHO grade I, and their postoperative clinical states showed a favorable progression according to the Karnofsky and McCormick scales.

Conclusion: WHO grade I histopathology and thoracic spinal location are predominant in SMs. With low recurrence and mortality rates, favorable outcomes can be obtained with gross total resection in most cases. Due to the slow course of SMs, care should be taken against the possibility of misdiagnosis.

Keywords: Histopathology; surgical approach; spinal meningioma

INTRODUCTION

Spinal meningiomas (SMs) are favorable lesions from an oncological surgical point of view (1). Meningiomas originating from arachnoid cells constitute 13-19% of all intracranial tumors. SMs, which frequently have an intradural extramedullary localization, constitute approximately 25-46% of all intradural spinal tumors, and their frequency among all meningiomas is 12% (2-6).

Typically, SMs are tumors that grow slowly and laterally to the subarachnoid space, have an intradural extramedullary localization, and begin to manifest clinical symptoms when there is compression of the spinal cord (4,7). Extracranial meningioma metastasis is exceedingly unusual, occurring in just around 0.1% of meningiomas (8). Completely extradural meningiomas are extremely rare (9). Approximately 80% of all SMs are seen in the thoracic region (2,10). They are generally non-invasive, well-circumscribed tumors that usually do not seed other parts of the central nervous system (11,12).

SMs are slow-growing benign tumors producing painless neurological deficits that often heal after surgery (4). The gold standard method in diagnosis is magnetic resonance imaging (MRI) (13,14). Meningiomas are mostly seen in the middle age group. Although cranial meningiomas are more common in women at a ratio of 2:1, this ratio is 9:1 for SMs (2).

SMs grow slowly out of intradural attachments, stretching and sometimes covering the arachnoid, but they rarely involve the pia mater (4,15). It is known that approximately 50% of SMs manifest with pain, and they have very variable clinical reflections including motor and sensory deficits and radicular pain (9,16). Long tract findings and Brown-Sequard syndrome may develop in the later stages of spinal cord compression (9). Very common changes in signs and symptoms may cause delays in diagnosis (9). Timely surgery plays an important role, especially in terms of postoperative functional outcomes. Sometimes satisfactory results can be obtained even in the presence of severe neurological dysfunction before surgery (10).

There are very few publications describing the postoperative results of SMs (9,10,15). Although intracranial meningiomas have been studied extensively from all aspects, there are only limited data concerning

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SMs. This study aimed to present the results of our series in terms of the natural course of SMs, surgical approach options, and pathology results in a comparative manner by examining the related data published in the literature.

MATERIALS and METHODS

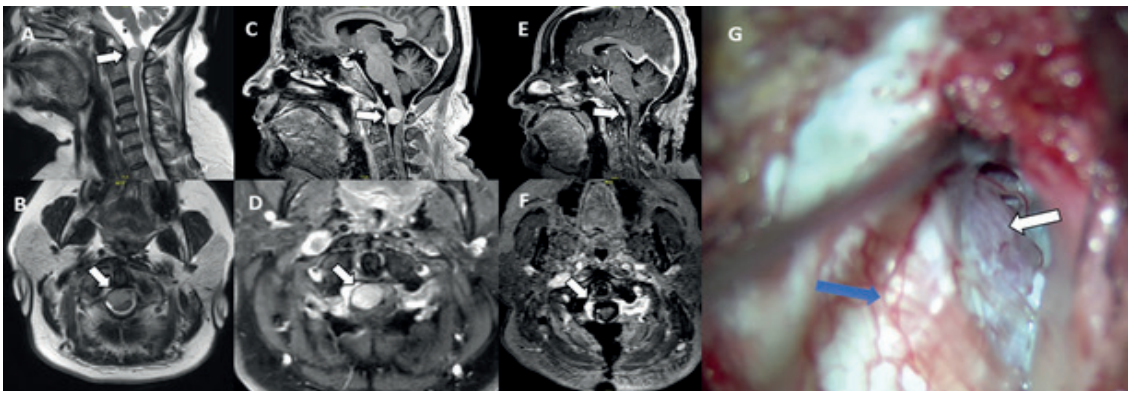
Of the 151 meningioma cases operated on between June 2014 and August 2020 in our center, 11 with spinal localization were analyzed retrospectively. Cases with foramen magnum and multiple meningiomas were not included in the study. The patients' age, gender, preoperative clinical findings, tumor level, histopathology, preoperative and postoperative third-month Karnofsky and McCormick scores are presented in Table 1. The study was conducted with the approval of the Ethics Committee of Istanbul Medipol University (E-10840098-772.02-66583).

RESULTS

Three (27%) of our cases were male and eight (73%) were female, and their mean age at the time of surgery was 58.9 (range; 21-75) years. Of all the meningioma cases, 7.2% (n = 11) were located in the spinal column. The mean follow-up period was mean 35 months (range; 6 – 74 months) in which we encountered no recurrence. It was observed that due to the slow clinical course of the tumor and the possibility of misdiagnosis, lumbar surgery had been recommended to two patients approximately six months before the operation. One of these patients had lumbar stabilization and decompression surgery but did not have any clinical benefit while the other did not accept the recommended surgery but presented to our center after developing sudden paraparesis.

Clinical symptoms and findings were especially in lower extremities quadriparesis in a C1-localized case (n = 1, 9%) (Figure 1), cervical pain in a C5-7-localized case (n = 1, 9%), and numbness in the right arm in a C3-localized case (n = 1, 9%). The remaining patients had varying degrees of paraparesis (n = 8, 73%).

Concerning tumor localization, three (27%) were observed at cervical level, eight (73%) at thoracic level (Figure 2). Anterior localization was seen in one tumor (9%), lateral in seven (64%), and posterior in three (27%). Total laminectomy was performed in a single case (9%) (Case 8) while the remaining cases underwent hemilaminectomy or laminoplasty.



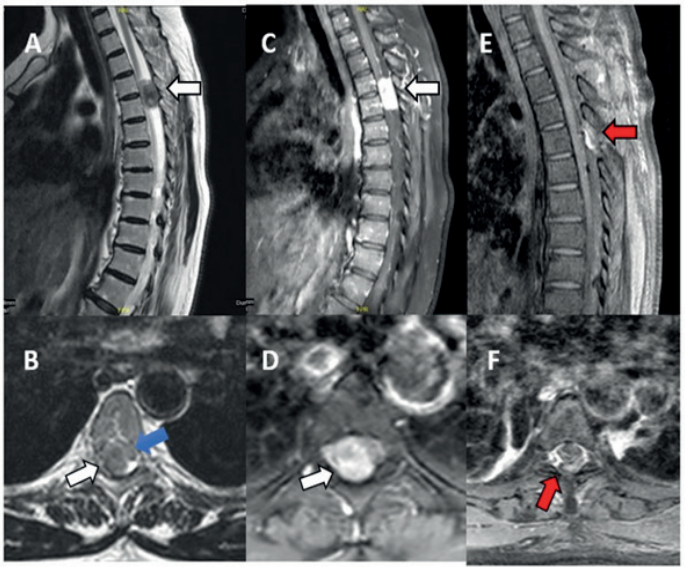
A,B: Preoperative T2-weighted, C,D: Preoperative contrasted T1-weighted, E,F: Postoperative contrast-enhanced T1-weighted, G: Peroperative (white arrows show the tumor and the blue arrow shows the spinal cord)

Figure 1. MRI images of Case 8

Table 1. Demographical aspects of our study

Case No	Age Gender	Clinical Manifestation	Level	Simpson Grade	Pathology (WHO, 2016)	Karnofsky Score (Preop/ Postop)	McCormick Score (Preop/ Postop)	Note
1	70/M	Paraparesis	T4-5	S1	Meningothelial	60/80	III/II	
2	75/F	Paraparesis	T2-3	S1	Metaplastic	50/80	III/II	*
3	47/F	Paraparesis	T4-5	S1	Psammomatous	60/90	III/II	Figure 2
4	65/F	Paraparesis	T4-5	S1	Transsitional	50/80	IV/II	**
5	65/M	Rightsided upper extremity numbness	C3	S1	Transsitional	60/90	III/I	
6	55/F	Neck pain	C5-7	S1	Transsitional	60/90	II/I	
7	21/M	Paraparesis	T5-6	S1	Psammomatous	50/80	III/II	
8	57/F	Quadriparesis	C1	S1	Meningothelial	50/90	III/II	Figure 1
9	73/F	Paraparesis	T4-5	S3	Meningothelial	50/60	III/III	
10	64/F	Paraparesis	T10	S1	Meningothelial	60/90	II/I	
11	56/F	Paraparesis	T5-6	S1	Transsitional	50/60	III/II	

M: Male, F: Female, T: Thoracal, C: Cervical, S: Simpson,
*: Lumbar stabilization, 6 months before surgery; **: 6 months before surgery L3-4 spinal stenosis decompression offer



A,B: Preoperative T2-weighted, C,D: Preoperative contrast-enhanced T1-weighted, E,F: Postoperative contrast-enhanced T1-weighted (white arrows show the tumor, the blue arrow shows the spinal cord, and red arrows show the operation area)

Figure 2. MRI images of Case 3

According to the 2016 WHO classification, all tumors were grade I, and the histopathological subtype was reported to be meningothelial in four cases (36%), transitional in four (36%), psammomatous in two (18%), and metaplastic in one (9%) (Table 1).

According to the preoperative and postoperative third-month Karnofsky and McCormick scale scores, all our cases benefited from surgery. The mean preoperative Karnofsky scale score was 54.5 (range; 50-60) while it was 80.9 (range; 60-90) at the postoperative third month. We did not encounter any intraoperative and early postoperative mortality. However, one of our cases died at the postoperative fourth month for another reason (abundant gastrointestinal bleeding).

DISCUSSION

SMs, which are considered as favorable tumors from an oncological surgical point of view, constitute 25-46% of all spinal tumors and are often localized in the thoracic region (1,5,6,10,17,18). Those located in the cervical and thoracal region are generally detected in the anterior and posterior of the spinal cord, respectively (2). SMs usually have an intradural extramedullary localization, and they are only rarely observed in the form of an epidural tumor, a tumor with an extradural extension, vertebral meningioma, or multiple spinal meningiomas (19-24). SMs are generally well-circumscribed, distinct dural-based tumors that are separated from the spinal cord and roots due to the arachnoid layer and are not actually considered to be true encapsulated tumors. Seventy-three percent of our SMs were located in the thoracic spine, and 90% had a posterior and lateral localization.

Clinical findings are progressive and very variable, including loss of strength, sensory changes, and radicular pain (9). In advanced stages of spinal cord compression, they can cause peripheral tumor growth, long tract sign, and Brown-Sequard syndrome. This heterogeneous nature of symptoms and findings may make it difficult to achieve a timely diagnosis (9). In today's technology, the gold standard diagnostic method is MRI. While computed tomography provides better information in cases of bone deformity, intra-tumor calcifications can also be seen, especially in SMs of the psammomatous type (25). Recently, Ono et al. emphasized that the tumor hardness due to calcification which may be related to preoperative neurological impairment could be defined heterogeneous contrast-induced T1-weighted MRI and CT findings (14). Among our cases, paraparesis (73%) was predominant.

The two times higher incidence of meningiomas in women compared to men may be related to various reasons such as the growth rate of the tumor being higher during pregnancy and in the presence of breast cancer, and this can be explained by the high rate of estrogen and progesterone receptors in meningiomas observed in women and the absence of female predisposition in children. Concerning the female predominance of these tumors, in addition to sex hormone receptors, steroid, peptidergic, aminergic and growth factor receptors also play a role (7,10). There was an 8:3 female predominance in our series.

The ideal surgical approach depends on the location of the tumor. Generally, one or two levels of laminectomy/laminoplasty or hemilaminectomy is sufficient for dorsal and dorsolateral tumors while for ventral or ventrolateral located tumors, more lateral expansion may be required, including costotransversectomy and/or partial vertebrectomy (26). Spinal instrumentation should also be kept in mind according to the width of the planned surgical field. Although ideal surgery is total resection, it has been reported that poor functional results may be obtained after surgery in calcified tumors due to their adhesion to the spinal cord and SM decompensation in multiple SMs (2,23,27,28). There is no case in our series clinically worsened.

Traditionally, the surgery of the dural segment to which isolated intradural SMs are attached involve the total resection of the tumor with the associated dura mater and the coagulation of the dural part forming the base of the tumor. Saito T. et al. described a different surgical technique, in which they preserved the outer layer of the dura mater (29). Although we were able to apply subtotal resection (STR) in one case with anterior spinal location (Case 9) in our series, we also achieved Simpson grade I [gross total resection (GTR)] in our remaining patients.

While the most important factor for local recurrence in SMs is subtotal resection, this rate has been reported as 1.3-6.4% over a period of one to 17 years (7,18,30). Hereby, Voldrich et al. reported 84 SM cases resected Simpson II in a 20-year long with the 32 months mean follow-up period. Although tumor recurrence rate reported as 5% in the main group, this rate reported as 44% in a subgroup followed up for a longer period of 6 years. Furthermore,

they emphasized that if the follow-up period is long enough, the tumor recurrence after Simpson II resection may be much higher than previously thought, particularly in younger patients. So, they conclude that patients with SM should monitored for a long-term (31). Naito et al. examined retrospectively 35 patients with WHO grade I SM operated over the past 10 years and followed-up for at least 2 years after surgery. They achieved Simpson grade I or II resection in 31 of 35 cases (88.6%) with no local recurrence during follow-up (32). Moreover, we did not encounter mortality and recurrence releated with spinal pathology and/or its surgery, since may be according to low grade histopathologies and accessible locations of our cases, yet.

As with cranial meningiomas, most SMs are grade I, and their subtypes are psammomatous, meningothelial, and transitional in order of frequency (11,18,33-35). Less frequently, grade II meningiomas as clear-cell and chordoid subtypes and grade III, anaplastic meningiomas with a higher risk of local recurrence have also been reported in the literature (12). Recently, Mauri et al. reported that for recurrence of SMs, Ki-67 index and arachnoid invasion are the risk factors, whereas tumor size, dural resection and progesterone receptor expression are not significant (36).

Prognostic factors in SM patients with severe motor deficits include the spinal cord level of the tumor, its relationship with the cord, degree of resection, radiologic features of tumor, preoperative neurological status, and surgical timing (2,10,14,15,31,36). In a series of 33 cases over a period of 15 years, it was determined that 66% of the cases were psammomatous, 22% were fibroblastic, and 11% were meningothelial in terms of histopathology. The intradural meningiomas had a lateral localization in 58% of cases, posterior in 24%, and anterior in 18%. Postoperative recovery was seen in 79% of the patients while the remaining 21% were observed to have worsened. The authors concluded that patients with meningiomas located in the posterior and lateral spinal canal and to the distal of the C4 level, those under 60 years of age, and those with a shorter preoperative symptom duration had better outcomes while patients presenting with psammomatous pathologies had less acceptable outcomes compared to the remaining variants (4).

In a systematic review of publications published until 2018, Pereira et al. examined 1829 meningiomas in a total of 1811 patients described in 24 articles. The mean age of 1450 female/361 male (4:1) cases was found to be 57.6 years, and it was reported that SMs were mostly located in the thoracic region (64.6%), followed by the cervical region (22.7%). In the same study, of the 1415 (83.3%) cases with histopathological data, 94.8% were grade I, 4.4% were grade II, and 0.8% were grade III, with the psammomatous (27.8%) and meningothelial (25.2%) variants being predominant. Among the 1454 cases for which data on resection limits were available according to the Simpson classification, GTR was performed in 94.5% (Simpson grade I and II) and subtotal resection in 5.5% (Simpson grade III or above). The authors also reported that the tumor recurrence rate was 4.4% (in 75 of 1722 cases) and the mortality rate based on disease progression or surgery was 3% (17).

There is no specific classification for SMs for the preoperative and postoperative evaluation of the neurological findings of the patients. However, McCormick's classification used in the literature for spinal ependymomas or other functional neurological classifications can also be employed for SMs (37,38). Parallel to technological developments, advances in imaging methods, neuroanesthesia and microsurgical techniques, and also application of preoperative embolization, peroperative ultrasonography, CUSA, technical developed surgical microscopes and intraoperative neuromonitoring methods lead to a rapid decrease in postoperative morbidity and mortality rates (7,11).

CONCLUSION

SMs are generally located in the thoracic region and are mostly WHO grade I histopathology tumors. GTR is possible with low recurrence and mortality rates, confirming the benign nature of most SMs. Due to the slow course of these tumors, care should be taken against the possibility of misdiagnosis, and a detailed clinical evaluation should be performed in all patients that are candidates for spinal surgery.

Competing Interests: The authors declare that they have no competing interest.

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REFERENCES

1. Raco A, Pesce A, Toccaceli G, et al. Factors Leading to a Poor Functional Outcome in Spinal Meningioma Surgery: Remarks on 173 Cases. Neurosurgery 2017;80:602-9.
2. Levy WJ. Bay J, Dohn D. Spinal cord meningioma. J Neurosurg 1982;57:804-12.
3. Parry DM, Eldridge R, Kaiser-Kupfer MI, et al. Neurofibromatosis 2 (NF2): Clinical characteristics of 63 affected individuals and clinical evidence for heterogeneity. Am J Med Genet 1994;52:450-61.
4. Schaller B. Spinal meningioma: relationship between histological subtypes and surgical outcome? Journal of Neuro-Oncology 2005;75:157-61.
5. Helseth A, Mork SJ. Primary intraspinal neoplasms in Norway, 1955 to 1986. A population-based survey of 467 patients. J Neurosurg 1989;71:842-5.
6. Kwee LE, Harhangi BS, Ponne GA, et al. Spinal meningiomas: Treatment outcome and long-term follow-up. Clin Neurol Neurosurg 2020;198:106238.
7. Gottfried ON, Gluf W, Quinones-Hinojosa A, et al. Spinal meningiomas: surgical management and outcome. Neurosurg Focus 2003;14:e2.

8. Sung CW, Hsieh KLC, Kuo YJ. A primary meningioma of the lumbar spine with neck metastasis. J Spinal Cord Med 2020;43:888-91.
9. Hollis PH, Malis LI, Zappulla RA: Neurological deterioration after lumbar puncture below complete spinal subarachnoid block. J Neurosurg 1986;64:253–6.
10. Solero CL, Fornari M, Giombini S, et al. Spinal Meningiomas: review of 174 operated cases. Neurosurg 1989;125:153-60.
11. McCormick PC, Post KD, Stein BM. Intradural extramedullary tumors in adults. Neurosurg Clin N Am 1990;1:591-608.
12. Saraceni C, Harrop JS. Spinal meningioma: chronicles of contemporary neurosurgical diagnosis and management. Clin Neurol Neurosurg 2009;111:221-6.
13. Schroth G, Thron A, Guhl L, et al. Magnetic resonance imaging of spinal meningiomas and neurinomas. Improvement of imaging by paramagnetic contrast enhancement. J Neurosurg 1987;66:695-700.
14. Ono K, Shimizu T, Fujibayashi S, et al. Predictive value of heterogeneously enhanced magnetic resonance imaging findings with computed tomography evidence of calcification for severe motor deficits in spinal meningioma. Neurospine 2020.
15. Ciapetta P, Domenicuci M, Raco A. Spinal meningiomas: prognosis and recovery factors in 22 cases with severe motor deficits. Acta Neurol Scand 1988;77:27-30.
16. Pansini A, LoRe F, Conti P, et al. Spinal Meningiomas. Springer, Berlin, 1996;1-242.
17. Pereira BJA, Almeida AN, Paiva WS, et al. Neuro-oncological features of spinal meningiomas: systematic review. Neurochirurgie 2020;66:41-4.
18. Gezen F, Kahraman S, Canakci Z, et al. Review of 36 cases of spinal cord meningioma. Spine 2000;25:727-31.
19. Cohen-Gadol AA, Zikel OF, Koch CA, et al. Spinal meningiomas in patients younger than 50 years of age: A 21-year experience. J Neurosurg 2003;98:258-63.
20. Frank BL, Harrop JS, Hanna A, et al. Cervical extradural meningioma: Case report and literature review. J Spinal Cord Med 2008;31:302-5.
21. Palmer JD, Cook PL, Ellison DW. Extracranial osseous metastases from intracranial meningioma. Br J Neurosurg 1994;8:215-8.
22. Rutherford SA, Linton KM, Durnian JM, et al. Epidural meningioma of the sacral canal. J Neurosurg Spine 2006;4:71-4.
23. Giammattei L, Penet N, Padovan S, et al. Decompensation of a thoracic meningioma below the operated level: A dramatic and unexpected complication. World Neurosurg 2020;140:162-5.
24. Pommier B, Vasiljevic A, Barrey C. Purely intraforaminal meningioma in the cervical spine. World Neurosurg 2021;145:432-3.
25. Lee JW, Lee IS, Choi KU, et al. CT and MRI findings of calcified spinal meningiomas: Correlation with pathological findings. Skeletal Radiol 2010;39:345-52.
26. Maiti TK, Bir SC, Patra DP, et al. Spinal meningiomas: clinicoradiological factors predicting recurrence and functional outcome. Neurosurg Focus 2016;41:E6.
27. Freidberg SR. Removal of an ossified ventral thoracic meningioma. Case report. J Neurosurg.1972;37:728-30.
28. Sandalcioglu IE, Hunold A, Muller O, et al. Spinal meningiomas: critical review of 131 surgically treated patients. Eur Spine J. 2008;17:1035-41.
29. Saito T, Arizono T, Maeda T, et al. A Novel technique for surgical resection of spinal meningioma. Spine. 2001;26:1805-8.
30. King AT, Sharr MM, Gullan RW, et al. Spinal meningiomas: a 20-year review. Br J Neurosurg 1998;12:521-6.
31. Voldrich R, Netuka D, Benes V. Spinal meningiomas: is Simpson grade II resection radical enough? Acta Neurochir (Wien). 2020;162:1401-8
32. Naito K, Yamagata T, Arima H, Takami T. Low recurrence after Simpson grade II resection of spinal benign meningiomas in a single-institute 10-year retrospective study. J Clin Neurosci 2020;77:168-174.
33. Sayagues JM, Tabernero MD, Maillo A, et al. Microarray-based analysis of spinal versus intracranial meningiomas: different clinical, biological, and genetic characteristics associated with distinct patterns of gene expression. J Neuropathol Exp Neurol. 2006;65:445-54.
34. Traul DE, Shaffrey ME, Schiff D. Part I: spinal-cord neoplasms-intradural neoplasms. Lancet Oncol 2007;8:35-45.
35. Lee JH, Sade B, Choi E, et al. Meningothelioma as the predominant histological subtype of midline skull base and spinal meningioma. J Neurosurg 2006;105:60-4.
36. Mauri F, De Caro MDB, Divitis O et al. Recurrence of spinal meningiomas: analysis oft he risk factors. Br J Neurosurg 2020;34:569-74.
37. McCormick PC, Torres R, Post KD, et al. Intramedullary ependymoma of the spinal cord. J Neurosurg 1990;72:523-32.
38. Setzer M, Vatter H, Marquardt G, et al. Management of spinal meningiomas: Surgical results and a review of the literature. Neurosurg Focus 2007;23:E14