

## MULTIPLE SPINAL CANAL MENINGIOMAS

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### ABSTRACT

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#### BACKGROUND

Meningiomas of the spinal canal are common tumours with the incidence of 25 percent of all spinal cord tumours. But multiple spinal canal meningiomas are rare in compare to solitary lesions and account for 2 to 3.5% of all spinal meningiomas. Most of the reported cases are both intra cranial and spinal. Exclusive involvement of the spinal canal by multiple meningiomas are very rare. We could find only sixteen cases in the literature to the best of our knowledge. Exclusive multiple spinal canal meningiomas occurring in the first two decades of life are seldom reported in the literature. We are presenting a case of multiple spinal canal meningiomas in a young patient of 17 years, who was earlier operated for single lesion. We analysed the literature, with illustration of our case.

#### MATERIALS AND METHODS

In September 2016, we performed a literature search for multiple spinal canal meningiomas involving exclusively the spinal canal with no limitation for language and publication date. The search was conducted through <http://pubmed.com>, a well-known worldwide internet medical address. To the best of our knowledge, we could find only sixteen cases of multiple meningiomas exclusively confined to the spinal canal. Exclusive multiple spinal canal meningiomas occurring in the first two decades of life are seldom reported in the literature. We are presenting a case of multiple spinal canal meningiomas in a young patient of 17 years, who was earlier operated for solitary intradural extra medullary spinal canal meningioma at D4-D6 level, again presented with spastic quadriplegia of two years duration and MRI whole spine demonstrated multiple intradural extra medullary lesions, which were excised completely and the histopathological diagnosis was transitional meningioma.

#### RESULTS

Patient recovered from his weakness and sensory symptoms gradually and bladder and bowel symptoms improved gradually over a period of two to three weeks.

#### CONCLUSION

Multiple spinal canal meningiomas confined exclusively to the spinal canal are very rare and no case was reported in the first two decades of life till now and this is the first case we are presenting and these can be excised safely with good postoperative recovery.

#### KEYWORDS

Multiple, Meningiomas, Spinal Canal.

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#### BACKGROUND

Spinal canal meningiomas account for 25 percent of all spinal cord tumors.<sup>1</sup> and usually they are solitary. Multiple spinal meningiomas are rare in compare to solitary lesions and account for 2 to 3.5% of all spinal meningiomas.<sup>2,3</sup> Multiple meningiomas defined as simultaneous or sequential appearance of 2 or more independently situated meningiomas of any histopathological type.<sup>4</sup> Most of the

reported cases of multiple spinal meningiomas are both intra cranial and spinal.<sup>1</sup> Exclusive involvement of the spinal canal by multiple meningiomas are very rare.<sup>1,5</sup> To the best of our knowledge we could find only sixteen case series were reported in the literature.<sup>2,3,6-19</sup>

Exclusive multiple spinal canal meningiomas occurring in the first two decades of life are seldom reported in the literature. We are presenting a case of multiple spinal canal meningiomas in a young patient of 17 years, who was earlier operated for single lesion. We analysed the literature with illustration of our case.

#### AIMS AND OBJECTIVES

To analyse all the reported cases of multiple meningiomas exclusively involving the spinal canal with illustration of our case. To analyse the aetiopathogenesis and results of multiple spinal canal meningiomas.

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## MATERIALS AND METHODS

In September 2016, we performed a literature search for multiple spinal canal meningiomas involving exclusively the spinal canal with no limitation for language and publication date. The search was conducted through <http://pubmed.com>, a well-known worldwide internet medical address. To the best of our knowledge, we could find only sixteen cases of multiple meningiomas exclusively confined to the spinal canal.

Multiple meningiomas exclusively involving the spinal canal are included in the study irrespective of age and sex. Multiple meningiomas involving the different neuroaxis are excluded in the study.

Exclusive multiple spinal canal meningiomas occurring in the first two decades of life are seldom reported in the literature. We are presenting a case of multiple spinal canal meningiomas in a young patient of 17 years, who was earlier operated for solitary intradural extra medullary spinal canal meningioma at D4-D6 level, again presented with spastic quadripareisis of two years duration and MRI whole spine demonstrated multiple intradural extra medullary lesions, which were excised completely and the histopathological diagnosis was transitional meningioma.

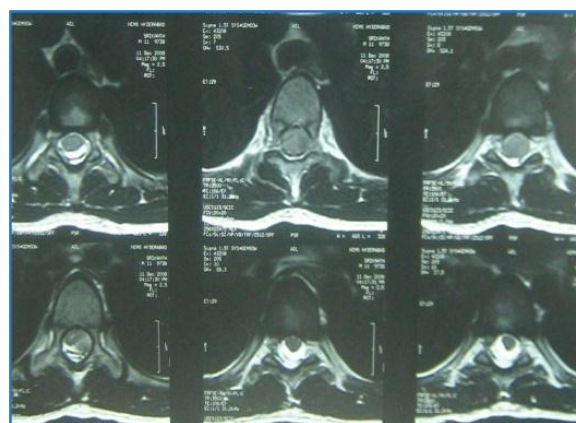
## Illustrated Case

In 2008, an eleven-year-old male patient presented with progressive weakness of both lower limbs of 2 months' duration with paresthesia. On neurological examination demonstrated spastic paraparesis with, decreased sensations below the D4 level with extensor plantars. On general examination there was no evidence of neurofibromatosis stigmata. MRI dorsal spine revealed intradural extra medullary lesion extending from D4-D6 with cord compression. (Figure 1 and Figure 2). D4-D6 laminectomy and excision of tumor was done. Total tumor was resected without any difficulty.

Histopathologically confirmed as psammomatous meningioma. Patient improved postoperatively and he was alright for 4 years and at the age of 17 years, patient again presented with two years history of weakness of all four limbs with decreased sensation below the clavicle with bladder and bowel disturbances. On clinical examination demonstrated quadripareisis with power of 3/5 with sensory deficit below C7. MRI whole spine demonstrated multiple intra dural extramedullary lesions at C2-C5, D3-D5, D6, D8, and at D10, likely of multiple meningiomas (Figure 3). Laminectomy from C2-C6, dorsal laminectomy D3-D6 and from D8-D11 was done with total excision of all the lesions were done. Histopathologically confirmed as transitional meningioma.



**Figure 1. Intradural Extra Medullary Tumor Extending from D4-D6**



**Figure 2. Intra Dural Extramedullary Tumor on the Lateral Aspect**



**Figure 3. Multiple contrast enhanced Intradural Extramedullary Lesions at C2-C5, D3-D5, D6, D8 and at D10**

## RESULTS

All the reported case series had good post-operative results, because these can be dissected freely from the cord and can be excised totally. Post-operative complications are transitory.<sup>19</sup> In our case, patient recovered from his weakness and sensory symptoms gradually and bladder and bowel symptoms improved gradually over a period of two to three weeks.

### Total list of Cases Published in the Literature till now

No	Year	Author	No of tumors
1	1952	Rand RW	1
2	1961	Lombadri G	2
3	1967	Rath S	1
4	1971	Scott M	1
5	1979	Puljic S	3
6	1982	Lewy` WJ	2
7	1983	Carta F	2
8	1984	DeRocco C	1
9	1989	Solero CL	1
10	1990	Weil SM	1
11	1991	Deda H	1
12	1993	Chaparro MJ	47
13	1993	Makiuchi T	1
14	1999	Lee KS	1
15	2003	Colazza GB,	1
16	2005	Silva JA	23
17	2016	Present case	1

## DISCUSSION

Anfimow and Blumenau first described multiple meningiomas and Cushing and Eisenhardt classified them as a distinct clinical entity.<sup>4</sup> Spinal meningiomas account for 25 percent of all spinal cord tumours and usually they are solitary.<sup>1,4</sup> Multiple spinal meningiomas are rare in compare to solitary lesions and account for 2 to 3.5% of all spinal meningiomas.<sup>2,3,4</sup> Multiple meningiomas defined as simultaneous or sequential appearance of 2 or more independently situated meningiomas of any histopathological type.<sup>4</sup> Most of the reported cases of multiple spinal meningiomas are both intra cranial and spinal.<sup>1</sup> Exclusive involvement of the spinal canal by multiple meningiomas are very rare.<sup>1,5</sup> Silva,<sup>19</sup> et al. reported a patient with 23 tumours of spinal canal meningiomas who was operated five times within 15 years period with good outcome. Chaparro,<sup>17</sup> et al. described a case with 47 distinctly separate spinal meningiomas. To the best of our knowledge we could find only sixteen cases in the literature. (Table).

Multiple spinal canal meningiomas are usually associated with von Recklinghausen's disease.<sup>1,4,8,12,19</sup> but these are reported without evidence of stigmata like our case.<sup>2,3,8,19</sup> Various theories have been put forward to explain the occurrence of multiple spinal canal meningiomas by Artunes and Zauk.<sup>20</sup>

The polycentric theory, the venous dissemination theory and the liquoric dissemination theory. Polycentric theory is widely accepted and according to this multiple spinal canal meningiomas are due to existence of multiple originating dural foci with predominant hereditary aspect.<sup>4,16,19</sup>

The pathogenesis of multiple spinal canal meningiomas can be explained in two ways, either these tumours arise independently as evidenced by the histological and cytogenetic differences between multiple tumours from the same patient, or a single transforming event occurs and the original clone of cells spreads throughout the meninges in the formation of multiple, clonally related tumours.<sup>21,22</sup> Multiple spinal canal meningiomas are reported following whole axis radiation for previous malignancy.<sup>23</sup> Colozza,<sup>18</sup> et al reported multiple spinal canal meningiomas in a 75 years female breast malignancy patient following tamoxifen therapy by saying the tumour inducing properties by the tamoxifen can responsible for the origin of multiple meningiomas.

Multiple spinal canal meningiomas are usually benign and commonly seen in adults with female dominance. Meningiomas occurring in children are having aggressive course with unfavourable prognosis.<sup>19</sup>

Multiple spinal canal meningiomas are not reported in the first two decades of life. We are the first one to illustrate the case of multiple spinal canal meningiomas in the 17 years male patient. We are presenting this case of 17 years old male, first operated at the age of eleven years for solitary meningioma in the thoracic level followed by asymptomatic period of four years. Again at the age of 17, presented with quadriparesis with multiple intradural extramedullary meningiomas in the thoracic level (4 tumours) and one in the cervical region, all the tumours were excised completely with good post-operative outcome.

Thoracic spine is most commonly involved followed by cervical and lumbar regions.<sup>19</sup> Meningiomas usually arise at the emergence of the sensitive root, in an intradural location, but rarely can occur in an epidural one. Rath,<sup>9</sup> et al. reported a case with intradural thoracic and extradural cervical meningiomas. Deda,<sup>16</sup> et al reported a case with meningotheial meningioma in the cervical region and fibroblastic meningioma in the lumbar region. Weil,<sup>15</sup> et al reported a case of intradural and extradural meningioma in the cervical spine cautioning intradural space should be evaluated carefully when an extradural meningioma is encountered.

The clinical picture depends on the involvement of the spinal level varying from radicular impairment to motor deficit with sensory and bladder and bowel involvement. MRI is the investigation of choice.<sup>2,19</sup> MRI whole spine with contrast examination usually gives the clear picture of multiple lesions and their radiological features are similar to the isolated meningiomas. An Intra dural extra medullary lesion with homogenous contrast enhancement is the typical finding in meningiomas. The differential diagnosis includes multiple neurofibromas, the combination of a meningioma and neurofibromas, "seed" metastases from

medulloblastomas and ependymomas and, rarely, extra CNS metastases.<sup>10</sup>

Surgery is the treatment of choice. All the tumours can be resected at the same time. These tumours grow slowly and these can be resected easily from the cord and dura. The post-operative complications are transitory. Silva.<sup>19</sup> et al. reported a case which was operated fifth time, that patient had paraplegia and urinary retention in the post-operative period which was improved gradually. Recurrences can involve the same level or at the different level, as in our case first time patient had solitary meningioma at thoracic level but multiple meningiomas occurred in the thoracic level and one tumour in the cervical level. Silva.<sup>19</sup> et al. reported a patient with 23 tumours of spinal canal meningiomas who was operated five times within 15 years period, all the tumours were in the thoracic spine at different levels.

Histology may be different. Most often these are psammomatous type, but other varieties were also reported. Scott.<sup>9</sup> et al. reported melanotic meningiomas. In our case first time histopathology was psammomatous meningioma and multiple lesions were transitional variety.

### CONCLUSION

Multiple spinal canal meningiomas exclusively involving the spinal canal are rare and occurring in the first two decades of life is not reported till to the date, and this is the first case in the literature we are presenting. These can be resected totally with good postoperative outcome.

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