

## Case Report

# Spastic Paraparesis in Young Men with Spinal Meningioma and Spinal Arteriovenous Malformation: A Case Report

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## ABSTRACT

**Background:** Compared to trauma cases, the publication of non-traumatic events is much less, although the incidence is estimated to be as much as a traumatic event. This case report presents spinal cord tumors together with arteriovenous malformation (AVM) in young men, where there are no previous case reports that the authors can find.

**Case Report:** A 22-year-old male patient with complaints of pain in the back (NPRS 8-9), followed by difficulty moving the lower limb accompanied by a burning sensation, problem in defecation, and urination. We found motor weakness in the lower extremities (1/1), positive pathological reflexes, and sensibility disorders on physical examination.

**Discussion:** According to history taking, physical examination, and MR Imaging, the spastic paraparesis symptom in this patient is likely due to a spinal cord tumor, which is meningioma (according to histopathological examination). Where back pain rarely presents in the patient with spinal AVM. Laminectomy and tumor destruction were the gold standards of therapy, and the symptom was relieved.

**Conclusion:** A complete examination is the main key in determining the appropriate therapy for the patient's disease. In this case, the manifestation of spastic paraparesis was likely to be caused by meningioma rather than spinal AVM. Laminectomy and extirpation of the spinal meningioma were done as the gold standard of therapy, and the symptom was relieved. In contrast, spinal AVM was not performed and is still under observation.

Keywords: Human and medicine; Thoracolumbar; Paraparesis; Meningioma; Arteriovenous-malformation

## INTRODUCTION

Spinal cord lesions are closely related to decreased quality of life.<sup>1,2</sup> Compared to trauma cases, the publication of non-traumatic events is much less, although the rate of occurrence is estimated to be as much as the traumatic event. In the future, it is estimated that the incidence of non-traumatic will increase; therefore, learning of the incidence, etiology, prevalence, and survival rate will be vital.<sup>3-6</sup>

The case report describes the incidence of extradural and intradural tumors with an arteriovenous malformation in young men, presenting with clinical gradual spastic paraparesis, urinating and defecating difficulties, and radiating back pain, where there are no previous case reports that the authors can find. The symptoms of spinal cord tumor and arteriovenous malformation are similar. The patient may present with progressive limb

weakness, complaints of urination, defecation, or sexual dysfunction. We hope to be a reference that can be used when the same case is present through this case report.

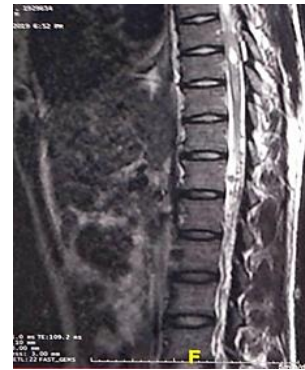
## CASE REPORT

The male patient, 22 years old, came with a chief complaint could not defecate and urinate for one week before admission. At first, the patient felt a localized pain in the back two months before hospitalization (Numerical Pain Rating Scale (NPRS) at admission 8-9); the pain was followed by the heaviness and weakness of lower extremities since one month before hospitalization, sensory disturbance also occurred the form of burning sensation. Weakness gradually felt from the sole to the entire extremities. The patient denies a history of trauma. The patient does not experience nausea, vomiting, headache, and vision problems.

In lower extremity motoric tests, 1 (right)/1 (left) motoric strength is obtained for anteflexion, retroflexion, abduction, upper limb adduction, lower leg flexion and extension, dorsiflexion and plantarflexion of the foot, and finger flexion and extension. On physiological reflex, there is no hyperreflexia. Babinski's examination found positive for both legs. Negative on sensibility examination as high as thoracal 6.

Cervicothoracolumbar vertebra MR Imaging was performed with T1, T2 sequences, without contrast. On MR Imaging obtained small hypointense lesions on T1 and hyperintense on T2 in the spinal canal as high

as the thoracal vertebra 9 (Figure 1-2), a picture of hypointense lesions on multiple T1 and T2 on the spinal canal along with the thoracal 11 to lumbar 5 (Figure 3).



**Figure 1.** Magnetic Resonance Image of thoracal and lumbar level, showing a hyperintense lesion in T2 weighted sagittal MR image at the level of thoracal vertebrae 9.



**Figure 2.** Hyperintense lesions in the spinal cord on the T2 axial section.



**Figure 3.** Multiple hypointense lesion in T2 weighted sagittal MR image in the spinal cord at the level of thoracal vertebrae 11 until lumbar vertebrae 5.

Laminectomy was done from vertebrae T-8 to vertebrae T-9, exposed an



extradural tumor with fibrous and vascularization, followed by the dura opening, which revealed a grey jelly-like mass and medulla. The extradural tumor was removed, and intradural mass was partly removed (0.5 cc). On histological examination of the tumor, the specimen showed meningioma. Microscopically, tumor tissue is arranged in islands, partly in bundles, monotonous cells with round - oval nuclei.

Postoperatively, the patient showed improvement of paraparesis and improvement in back pain. Five days after surgery, the patient was dismissed from the hospital with improvement in lower motoric function (3/3) and pain (NPRS 5). The patient's home medication was analgesic, gabapentin, and a supplement of B12 vitamin. Thirteen days after the surgery, the patient showed gradual improvement in the motor (4/3) and sensation disturbance. The patient also minimally feels the pain in his back (NPRS 2).

## DISCUSSION

### Spinal Meningioma

Spastic paraparesis is a characteristic of upper motor neuron lesions. Whereas it affects both of the legs, it comes from a spinal cord lesion. It can manifest acute injury, infection, intervertebral disc prolapsed, vertebral canal stenosis, spinal cord tumors, intrinsic cord lesion, or the least common arteriovenous malformation.<sup>7</sup>

Spinal cord tumor is 15% of central nervous system tumor.<sup>8</sup> The prevalence of primary spinal cord tumors is 4 - 8% of central

nervous system tumors.<sup>9</sup> Spinal cord tumors can be classified by their location: extradural, intradural extramedullary, and intradural intramedullary. The most common findings in patients with spinal cord tumors are pain, starting with localized pain and then radiates. Intramedullary tumor pain is characterized by atypical and diffuse pain.<sup>8</sup> The pain does not feel better, even with analgetics. There is also a sensory disturbance that was caused by the compression of the spinal cord. Other classical findings with spinal cord tumors are the motoric dysfunction that radiates according to the radix that was compressed by the tumor. There can be a urination and defecation problem. The patient can also complain about increasing intracranial pressure symptoms, like nausea, vomiting, headache, vision disturbance, etc. MRI and CT scans are the gold standards of examination in a spinal cord tumor.<sup>10</sup>

In this patient, the neurological symptoms progressed over two months. The first symptom in the patients was pain that radiates from the back pain through both legs; pain did not feel better, even with analgetics. The pain was followed by sensory disturbance. The patient also complained about the weakness of both legs that worsened until he cannot walk one month before admission. The patient could not urinate and defecate for one week before admission.

In the physical examination, the patient could not move his leg and feet. There is also a pathological reflex (Babinski reflex) and clonus on both legs. In the sensory examination, there was hypoesthesia in both legs. Magnetic Resonance Imaging showed a hypointense



lesion in the T1 sequence and a hyperintense lesion in the T2 sequence at the level of thoracal vertebrae 9 with homogenous enhancement with contrast. This lesion led us to a diagnosis of a spinal cord tumor.

Laminectomy was done from vertebrae T-8 to vertebrae T-9, exposed an extradural tumor with fibrous and vascularization, followed by the dura opening, which revealed a grey jelly-like mass and medulla. The extradural tumor was removed, and the intradural tumor was partly removed (0.5 cc). On histological examination of the tumor, the specimen showed meningioma. Microscopically, tumor tissue is arranged in islands, partly in bundles, monotonous cells with round - oval nuclei (meningothelial whorls).

According to *the Central Brain Tumor Registry of the United States*, meningioma was the most frequently reported brain and other Central Nervous System (CNS) tumor, accounting for 37.1% of the tumor. Only 4.2% of meningioma were located in the spinal meninges. Meningioma is most common in adults age 65 years and older.<sup>11</sup> Meningioma is more common in women rather than in men (meningioma has progesterone receptor). Meningioma is the most common extramedullary tumor. Sixty to seventy percent of meningioma was found in the thoracal level of vertebrae. Resection of meningioma has a high success rate (80-95%) with low recurrence (4-8%) with good clinical improvement.<sup>8,12</sup>

The patient did not show any significant risk factors. Several studies have shown the

association of genetic mutations neurofibromatosis with intramedullary spinal cord tumors that often occur at a young age. This association should be investigated further due to limitations in our facilities. The patient also did not find behavior that could increase the hormone progesterone excessively, although this would later have to be scientifically proven with investigations. Thus far, the risk factors for patients having tumors in the spinal cord have not been found.<sup>13</sup>

Laminectomy and extirpation of the tumor were done in this patient as the gold standard therapy for spinal cord tumors, with a high success rate (80-95%) and low recurrence rate (4-8%).<sup>8</sup> The patient has a good clinical improvement after the spinal cord tumor resection; there is an improvement in lower motoric function 13 days after removing the tumor (from 1/1 to 4/3) and back pain (NPRS 8-9 at admission to NPRS 2). However, due to the subtotal excision of the intradural tumor, further observations should be made in the next few years.

### Spinal Arteriovenous Malformation

On MRI examination, there are multiple hypointense lesions in the T1 and T2 sequence in the spinal cord at the level of thoracal vertebrae 11 until lumbar vertebrae 5. Multiple signal voids suggest a dural arteriovenous malformation at the level of thoracal vertebrae 11 until lumbar vertebrae 5.

Spinal arteriovenous malformations (AVMs) are relatively rare, constituting 3-4% of all spinal cord space-occupying lessons. The commonly used classification proposed by



Abson and Spezler divides spinal AVMs into four categories: type I, dural AVFs; type II, intramedullary glomus AVMs; type III, juvenile or combined AVMs; and type IV, intradural perimedullary AVF. Spinal AVM also can be classified according to its location: dural arteriovenous fistula, intramedullary AVM, and intradural perimedullary - subpial AVM.<sup>14,15</sup>

Spinal dural arteriovenous fistulas (DAFs) are the most common spinal vascular malformation, commonly affecting men, mostly occurring at 50-60 years. This type of AVM presents slowly progressive weakness of the lower extremity, back pain, or radicular pain. Type II, Intramedullary AVM is a congenital malformation commonly occurs within the thoracolumbar region, commonly present in young adults with an average age mid-20s. Type III Juvenile AVM often occurs at a younger age than type II. These two types often have the same picture with bleeding and myelopathy, which the involvement of surrounding tissues can distinguish. Type IV intradural perimedullary AVF is characterized by a shunt between the radicular artery and intradural veins, with an incidence of 4-17% of all AVMs.<sup>14,15</sup>

Spinal digital subtraction angiography (DSA) is the gold standard in AVM visualization, MRI compared to DSA, is still not good at assessing structures surrounding the lesion. The presence of cord edema accompanied by an increase in T2 signal and flow voids can direct the diagnosis to AVM. Other supports that can be used are Magnetic Resonance Angiography (MRA) and Computed Tomography Angiography (CTA) 15.<sup>14</sup>

Based on the history, physical examination, and MRI, the patient may have type III AVM. Further investigations and intraoperative findings must support the diagnosis of this type. The diagnosis is also complicated by the presence of meningiomas in the patient.

Management of spinal AVM can be divided based on its classification, and effective management can be surgery and endovascular treatment.<sup>11</sup> Patients with suspected type III can be observed first because of the possibility of new malformations appearing after drainage. The patient was also decided to treat the meningioma first and observe the AVM.<sup>15-17</sup>

According to history taking, physical examination, and MR Imaging, the spastic paraparesis symptom in this patient is likely due to a spinal cord tumor, which is meningioma (according to histopathological examination) due to the back pain that is rarely present in the patient with spinal AVM.

The patient has a good clinical improvement after the spinal cord tumor resection; there is an improvement in lower motoric function 13 days after the removal of the tumor (from 1/1 to 4/3) and back pain (NPRS 8-9 at admission to NPRS 2).

Currently, there are very few reports that report AVM cases and spinal meningiomas at the same time. Still, there have been several reports of events in the cerebral. With the small number of case findings, it is not certain whether the incidence of AVM and meningioma is related. There are several hypotheses. Cushing and Heisenhardt revealed that chronic phlogosis of the arachnoid cells with a typical



high flow rate on arteriovenous shunt of AVMs could be the key factor to facilitate the arising of benign brain tumors. Kasanticul et al. speculated that tumor angiogenesis factor (TAF) and other AVM-released cytokines create a favorable environment for tumor growth.<sup>18</sup>

## CONCLUSION

There are still very few published cases of finding spinal meningiomas and spinal AVMs that occur together. It is still unclear whether there is a relationship between the two events, although there are several hypotheses. A complete examination is the main key in determining the appropriate therapy for the patient's disease. In this case, the manifestation of spastic paraparesis was likely to be caused by meningioma rather than spinal AVM. The patient, in this case, is thought to have AVM type III, which must be proven by further examination. Laminectomy and extirpation of the spinal meningioma were done as the gold standard of therapy, and the symptom was relieved. In contrast, spinal AVM was not performed and is still under observation.

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