



# Long-Term Follow-Up of Coexisting Meningioma and Intramedullary Ependymoma as a Collision Tumor in the Spinal Cord: A Case Report

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## Abstract

**Background:** Meningioma and ependymoma are usually presented as intramedullary and extramedullary tumor of spinal cord, respectively. However, coexistence of meningioma and ependymoma as a single mixed tumor in the single spinal cord and canal is an extremely rare occurrence. To our knowledge, there have been no reports on a mixed tumor with a distinct meningioma in thoracic region and ependymoma occurring at the cauda equina. Here, we report a case of long-term follow-up of coexisting meningioma and intramedullary ependymoma as a collision tumor in the spinal cord.

**Case Presentation:** A 58-year-old woman presented with claudication in the lower limb one month after a fall from a step, characterized by weakness, numbness, and pain. MRI revealed two spinal lesions, one at the T4 level and the other in the cauda equina, at the L2 level, with L1 vertebral fracture. The patient underwent tumor resection *via* osteoplastic laminotomy from T3 to T5 and L2 to L3 simultaneously. Histopathological examination revealed that the tumor at the T4 level was a WHO grade I meningioma and that the lesion at L2 was a WHO grade II ependymoma. The postoperative course after 8 years was uneventful, with no recurrence, and the patient's symptoms resolved completely.

**Conclusion:** Meningioma and ependymoma represented separate and distinct fragments of the tumor that abutted one another, lending credence to the so-called "collision tumor in the spinal cord" theory. A careful diagnostic strategy should be considered for these multiple tumors in the whole body.

## Background

Coexistence of schwannoma and meningioma as a single mixed tumor is an extremely rare occurrence. To our knowledge, there have been no reports on a mixed tumor with a distinct meningioma in thoracic region and ependymoma occurring at the cauda equina. Here, we report a case of long-term follow-up of coexisting meningioma and intramedullary ependymoma as a collision tumor in the spinal cord. We had an informed consent to the patient for this case report.

## Case Presentation

A 58-year-old woman presented with claudication in the lower limb one month after a fall from a step, characterized by weakness, numbness, and pain. Sensory weakness was noted below the nipp. On neurological examination, bi-lateral Babinski test results were positive, and deep reflexes were also hyper bilaterally. There was no evidence of skin pigmentation abnormality and no family history of neurogenic tumors such as Neurofibromatosis type 2 (NF-2). The patient was previously diagnosed with a vertebral compression fracture at the L1 level.

Whole body Magnetic Resonance Imaging (MRI) revealed two spinal lesions, one of which was at the T4 level and the other in the cauda equina, at the L2 level, with L1 vertebral fracture. Gadolinium enhanced MRI showed ring enhancement of lesions that demonstrated intradural extramedullary tumors (Figure 1). Further, a homogenous enhanced lesion occurred at the L2 level, and it could not be determined whether the lesion originated from a single nerve root in the cauda equina (Figure

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Received Date: 02 Jan 2019

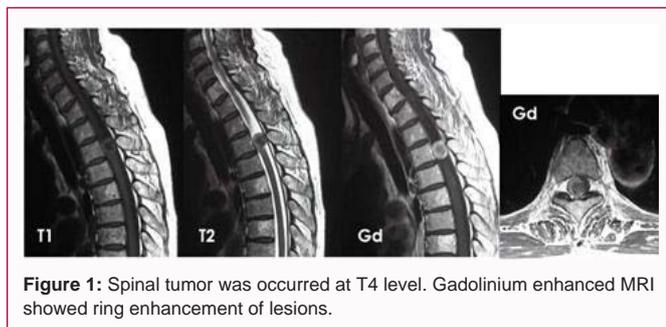
Accepted Date: 29 Jan 2019

Published Date: 31 Jan 2019

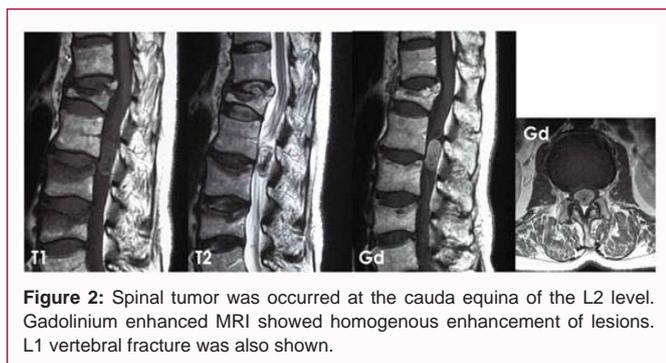
### Citation:

Teraguchi M, Kawakami M, Nakao S, Fukui D, Nakagawa Y. Long-Term Follow-Up of Coexisting Meningioma and Intramedullary Ependymoma as a Collision Tumor in the Spinal Cord: A Case Report. *Clin Surg*. 2019; 4: 2317.

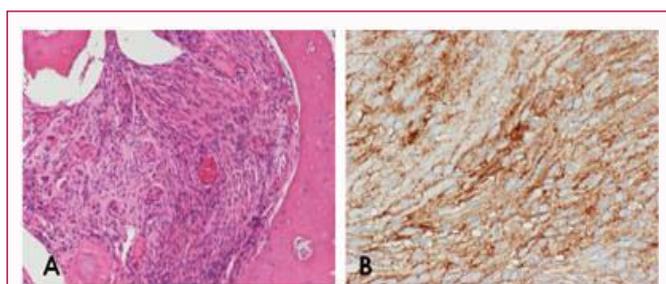
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**Figure 1:** Spinal tumor was occurred at T4 level. Gadolinium enhanced MRI showed ring enhancement of lesions.



**Figure 2:** Spinal tumor was occurred at the cauda equina of the L2 level. Gadolinium enhanced MRI showed homogenous enhancement of lesions. L1 vertebral fracture was also shown.



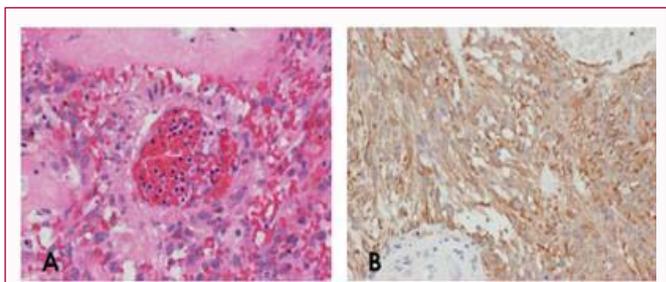
**Figure 3:** Meningioma component. (A) The tumor cells are characterized by clusters of epithelioid meningothelial cells with abundant eosinophilic cytoplasm and regular nuclei forming spindle shape (Hematoxylin-Eosin (HE) stain, x20), (B) positivity for epithelial membrane antigen (EMA, x40).

2). Computed Tomographic (CT) also demonstrated intradural extramedullary tumors, wherein calcification and ossification into the tumor at the T4 level were observed.

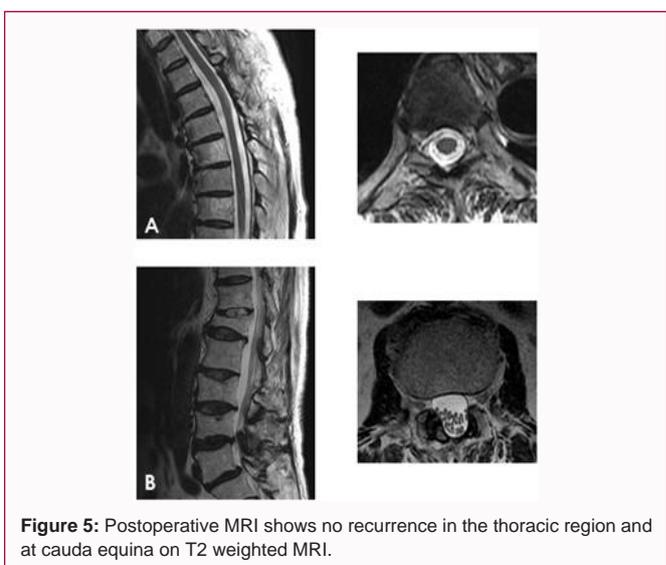
The patient underwent tumor resection via osteoplastic laminotomy from T3 to T5. After opening the dura, a 2.0 × 2.0 cm<sup>2</sup> reddish tumor was found at the T4 vertebral level, which was identified as originating from the arachnoid. Simultaneously, we resected the cauda equina tumor at L2 after partial laminectomy from L2 to L3. The appearance was consistent with that of an ependymoma. No significant atypia, mitotic activity, or necrosis was seen in these fragments.

Histopathological examination revealed that the tumor at the T4 level was a World Health Organization (WHO) grade I meningioma. The tumor cells are characterized by clusters of epithelioid meningothelial cells with abundant eosinophilic cytoplasm and regular spindle-shaped nuclei (Hematoxylin-Eosin (HE) stain (A), and positivity for Epithelial Membrane Antigen (EMA) (B) (Figure 3).

Further, examination of the cauda equina tumor revealed that it was a WHO grade II ependymoma. Histopathological examination



**Figure 4:** Ependymoma component: (A) Eosinophilic spindle shape cytoplasm with papillary and perivascular pseudorosette architecture (HE stain, x40), (B) The cells showed positive reaction for glial fibrillary acidic protein (GFAP) (Immunohistochemical stain, x200).



**Figure 5:** Postoperative MRI shows no recurrence in the thoracic region and at cauda equina on T2 weighted MRI.

showed that the cells had an eosinophilic spindle-shaped cytoplasm with papillary and perivascular pseudorosette architecture on HE staining (A) and that the cells showed a positive reaction for Glial Fibrillary Acidic Protein (GFAP) (B) (Figure 4).

Post-operatively, the patient fully recovered and was subsequently discharged. Post-operative CT and MRI revealed no sign of remnant tumor or recurrence. Because of complete tumor resection, the patient has been under monthly follow up to check for recurrence; thus, we did not perform radiotherapy. Furthermore, the postoperative course over 8 years has been uneventful, with no recurrence, and the patient’s symptoms have resolved completely (Figure 5).

### Discussion and Conclusion

Since the first report of mixed tumor occurrence in one spinal cord tissue by Cushing and Eisenhart in 1938 [1], only few cases of mixed schwannomas and meningiomas in both components seen together in patients with NF-2 have been published [2-6].

Even more rarely, to our knowledge, mixed schwannomas and meningiomas have been reported in patients without definite clinical evidence of NF-2 [7-9]. Rasheed et al. reported a mixed tumor comprising schwannoma admixed with meningioma and ependymoma in the cervical spinal cord in association without NF-2 [7]. There are only 3 case reports of such mixed tumors in patients without NF-2 included central verve system [7-9]. They proposed a theory on the occurrence of mixed components in the same tumor, according to which mixed tumors occur because of “collision” of

2 separate tumors that had originally developed independently at different times. Our patient, who had no symptoms or characteristics of neurofibromatosis as well as no family history of neurogenic tumors, had a mixed tumor with components of meningioma in the thoracic region and ependymoma at the cauda equina.

Some authors stipulate that this type of mixed tumor may result from reactive meningotheial proliferation adjacent to a schwannoma [5], whereas other reports hypothesize that a predominant tumor type contains areas that resemble other types of tumor [10]. Kim et al. proposed an alternative hypothesis, according to which mixed components in the same tumor result from differentiation from a common progenitor cell line [6]. However, the authors could not provide convincing evidence for common progenitor cells for Schwann cells, meningotheial cells, and ependymal cells. Therefore, the precise mechanism of the histogenesis of these mixed tumors remains unresolved, and currently, these tumors are reported as essential tumor combinations.

To our knowledge, this is the first report of a mixed tumor with components of a meningioma in the thoracic region and ependymoma at the cauda equina. The components of the meningioma and ependymoma represent separate and distinct fragments of the tumor that coexist one another, lending credence to the “collision tumor in the spinal cord” theory. Therefore, a careful diagnostic strategy should be considered for these multiple tumors in the whole body.

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