

# A Rare Case of Cervical Dumbbell Meningioma with the C2 Spinous Process Preservation

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Received 18 March 2016; accepted 18 April 2016; published 21 April 2016

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## Abstract

Dumbbell growth is often observed in schwannoma but rarely in meningioma. In particular, the occurrence of dumbbell meningioma at the cervical cord level in Japan is markedly rare. We reported an elderly case with symptomatic meningioma that developed in a dumbbell shape from cervical spine and was treated successfully by surgery.

## Keywords

Dumbbell Meningioma, Spinous Process Preservation, Cervical Spine, Spinal Tumor, Spinal Surgery

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## 1. Introduction

Intradural extramedullary tumors account for approximately 70% - 75% of spinal cord tumors, and meningioma and schwannoma account for approximately 70% of them [1] [2]. In Western countries, the occurrence of meningioma and schwannoma is comparable [2], whereas the occurrence of meningioma in Japan is approximately one-fourth of that of schwannoma [3]. Dumbbell growth is often observed in schwannoma, but rarely in meningioma. In particular, the occurrence of a dumbbell meningioma at the cervical cord level is markedly rare. Here, we reported an elderly case with symptomatic meningioma that developed and grew in a dumbbell shape at the spinal cord and was treated successfully by surgery.

## 2. Case Report

A 76-year-old female had complained of numbness in her right hand for 10 months and visited a nearby clinic

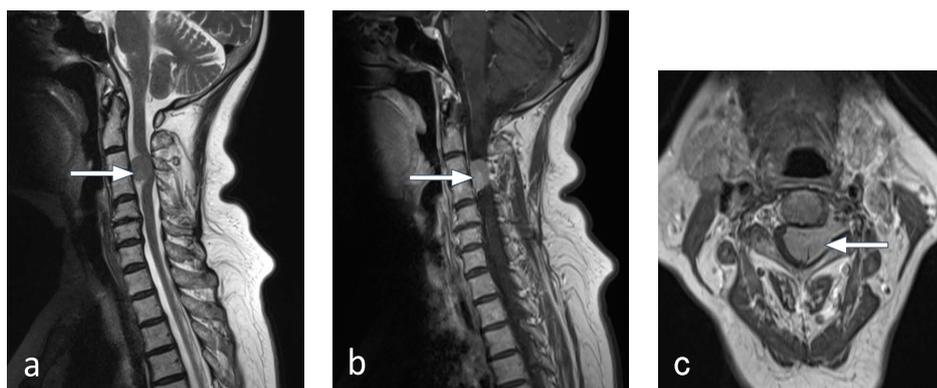
**How to cite this paper:** Sato, T., Okuda, T., Saito, T., Yonezawa, I., Onda, S., Shimamura, Y., Yoshikawa, K., Takano, H. and Kaneko, K. (2016) A Rare Case of Cervical Dumbbell Meningioma with the C2 Spinous Process Preservation. *Open Journal of Orthopedics*, 6, 98-102. <http://dx.doi.org/10.4236/ojo.2016.64014>

for rehabilitation. She was referred to our hospital for pain and weakness in the left upper and lower extremities, impaired coordinated movement of the left hand, and pain in the right lower extremity. Physical examinations revealed decreased muscle strength of the left upper and lower extremities, pain in the upper and lower extremities, palsy at the C5 region of the left upper extremity, coordination disturbance, bilateral hyper reflex of the biceps and its distal tendons, and spastic gait with a Japanese Orthopedic Association (JOA) score of 11 points.

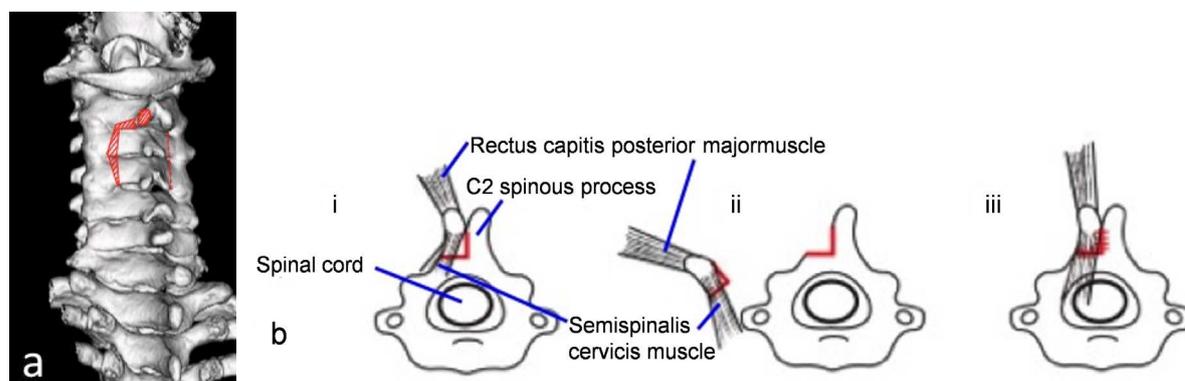
Magnetic resonance imaging (MRI) demonstrated a well-demarcated mass, 2 cm in size that exhibited slightly higher intensity on T2-weighted imaging, iso-intensity on T1-weighted imaging, and homogenous contrast enhancement with the dural tail sign on contrast imaging, which compressed the cervical cord. The mass, surrounding the vertebral artery, extended to the outer space of the left C3/4 intervertebral foramen and was categorized as a type-2 dumbbell tumor according to Eden's classification (**Figure 1**).

The patient underwent operation to excise the tumor. The C2 spinous process was half-dissected and inverted to the left with the rectus capitis posterior major muscle, oblique capitis inferior muscle, and semispinalis cervicis muscle still attached. Subsequently, the medial half of the facet joint on the left side of the vertebral arch was partially resected and a C3-4 half-opened vertebral arch was created. Then, the whole view of the extraforaminal part of the tumor, extending from the C2 tail to C4, was exposed (**Figure 2**).

First, the intratumoral pressure of the extradural section of the tumor was reduced. With regard to the anterior section of the tumor involving the vertebral artery, tumor enucleation was carried out as much as possible. At



**Figure 1.** Preoperative MRI. (a) A mass with slightly high intensity signals was observed at the C3/4 level by sagittal MRI on T2-weighted imaging (arrow); (b) Sagittal; and (c) Axial Gadolinium-enhanced T1-weighted images. A well-demarcated dumbbell mass, 2 cm in size, and homogeneously enhanced with the dural tail sign, compressed the cervical spinal cord at the C3/4 level. The mass, extending from the intradural space to the left C3/4 neural foramen in a dumbbell shape, was also present around the left vertebral artery (arrow).



**Figure 2.** Preoperative 3D CT. (a) The solid line indicates the dissected area, the shaded part shows the area of bone resection, and the dotted line indicates the hinge section; (b) A scheme showing the deep layer extensors at the cervical spine. 1) The left side of the spinous process was additionally half-incised with the C2 spinous process-attached muscles still attached; 2) It was possible to observe under direct vision the outside part of the tumor via the intervertebral foramen at the C3 and C4 levels by introversion of the C2 spinous process to the left lateral side; 3) The C2 spinous process was restored and attached to the right side of the spinous process for reconstruction.

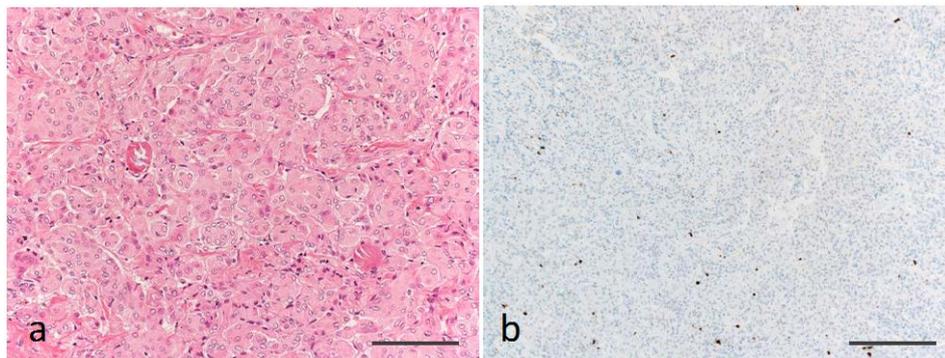
that time, intraoperative pathology consultation led to the diagnosis of meningioma.

Then, the whole intradural section of the tumor was resected with the dura mater, which was considered the site of primary occurrence, with debulking advanced using a Cavitron Ultrasonic Surgical Aspirator. The defective area of the dura mater was sutured with artificial dura mater, and the inverted C2 spinous process was returned and sutured with a non-absorbable thread. A cervical collar was attached for one month after surgery. The case was classified as Simpson grade IV. Pathologically, meningioma was diagnosed. MIB- I staining-positive cells accounted for 3% - 4% of all cells in each field of vision (Figure 3).

The postoperative clinical course has been uneventful for 2 years after the operation, and her some symptoms have been alleviated. The JOA score was 17 points and Hirabayashi's recovery rate was 100% [4]. Computed tomography (CT) at approximately 6 months after the operation revealed bony union at the C2 spinous process and preservation of the outside half of the C3/4 facet joint (Figure 4). MRI at 2 years after the operation demonstrated a residual tumor around the vertebral artery, but there was no apparent growth (Figure 5).

### 3. Discussion

To the best of our knowledge, only 8 cases with dumbbell meningioma developing at the cervical vertebral level have been reported to date [1] [5]-[11]. Schwannoma often develops into a dumbbell type. Meanwhile, meningioma develops from the arachnoid membrane and often adheres to, but rarely penetrates, the dura mater [12]. It grows along the inner side of the dura mater and is unlikely to develop into a dumbbell type. Therefore, the de-



**Figure 3.** Pathohistological findings. (a) H.E. staining (×200); (b) MIB-1 staining (×100). Scale bars = 100 μm (a), and 200 μm (b). Tumor cells consisted of an elliptical nucleus and eosinophilic cytoplasm, and psammoma bodies were also recognized. MIB-1 staining-positive cells accounted for 3% - 4% of all cells in each field of vision.



**Figure 4.** Postoperative 3D CT Synostosis was observed at the C2 spinous process.

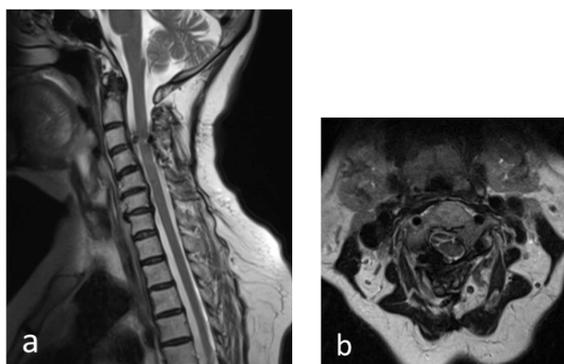
velopment of a dumbbell meningioma at the cervical vertebral level is markedly rare.

This is the sixth case report of dumbbell meningioma occurring at the cervical vertebral level and involving the vertebral artery [5]-[7] [9] [10] (Table 1). In all cases, the tumor was enucleated and resected only at Simpson grade IV [13]. The postoperative follow-up period showed a wide difference from 6 months to 5 years, but no case showed aggravation, such as recurrence. In our case, a residual tumor was observed around the vertebral artery. However, the JOA score was 17 points and Hirabayashi's recovery rate was 100%. In addition, Yoshihara *et al.* mentioned that it was acceptable to resect partially the extradural part of a benign tumor adjacent to the vertebral artery to avoid injury to the artery [14]. Mirimanoff *et al.* [15] reported that the 10-year tumor recurrence rate of spinal cord meningioma was 13%. Since the present case, at 76 years of age, was the oldest case described to date, the tumor around the vertebral artery was resected only as much as possible, with the risk of vertebral artery injury taken into account. Considering the result of our case, it is acceptable to resect partially the extradural part of low growth rate meningioma adjacent to the vertebral artery to avoid much risk in elderly cases.

Long-term symptomatic improvement is expected following complete resection of the intradural section of a tumor, including the dural occurrence site, and decompression of the cervical spinal cord. In the posterior approach for cases with a Eden's type-2 upper cervical dumbbell spinal tumor, inversion of the group of C2 spinous process-attached muscles with the spinous process still attached provides an excellent view of the C2/3 facet joint, including the caudal side, and enables maneuvers around the vertebral artery. In addition, reconstruction of C2 spinous process-attached muscles and bony union can be expected by returning the inverted C2 spinous process to its original position. Furthermore, postoperative vertebral instability and axial pain may be prevented.

#### 4. Conclusion

This is a rare case of dumbbell meningioma occurring at the cervical spinal cord in an elderly patient. Even when total resection of meningioma is unfeasible and an extradural residual tumor is recognized, tumor growth is often slow, and one option is not to adhere to total resection in elderly cases when operative invasiveness is taken



**Figure 5.** Postoperative MRI. (a) Sagittal and (b) axial T2-weighted images. A residual tumor was observed around the vertebral artery at the C3/4 level.

**Table 1.** Summary of cases with cervical dumbbell type meningioma in the literature.

Reference	Age (years), sex	Level	Simpson's grade	Postoperative residual tumor around the vertebral artery*	Recurrence**	Follow-up period
Ozaki <i>et al.</i> [5]	49, F	C2–4	IV	+	–	2.5 years
Hakuba <i>et al.</i> [6]	60, M	C7–T1	IV	+	–	5 years
Yoshiura <i>et al.</i> [7]	16, M	C2–4	IV	+	–	8 months
Chen <i>et al.</i> [9]	16, F	C1–2	IV	–	–	10 months
Clarke <i>et al.</i> [10]	19, M	C7–T1	IV	+	–	14 months
Current case	76, F	C3–4	IV	+	–	2 years

\*+: A tumor remained around the vertebral artery; -: A small piece of tumor remained over the posterior wall of the intervertebral foramen; \*\*: There was neither clinical nor radiologic evidence of tumor recurrence during the follow-up period.

into account. Securing an intraoperative visual field and operative maneuvers are easier by inversion of the group of C2 spinous process-attached muscles with the spinous process still attached, and returning it to its original position after tumor extraction facilitates reconstruction.

## Consent

The patients have given her informed consent for the case report to be published.

## References

- [1] Ozwa, H., Kokubun, S., Aizawa, T., Hoshikawa, T. and Kawahara, C. (2007) Spinal Dumbbell Tumors: An Analysis of a Series of 118 Cases. *Journal of Neurosurgery: Spine*, **7**, 587-593. <http://dx.doi.org/10.3171/SPI-07/12/587>
- [2] Simeone, F.A. (1975) Intraspinous Neoplasms. In: Rothman, R.H. and Simeone, F.A., Eds., *The Spine*, WB Saunders, Philadelphia, 35-42.
- [3] Jinnai, T., Hoshimaru, M. and Koyama, T. (2005) Clinical Characteristics of Spinal Nerve Sheath Tumors :Analysis of 149 Cases. *Neurosurgery*, **56**, 510-515. <http://dx.doi.org/10.1227/01.NEU.0000153752.59565.BB>
- [4] Hirabayashi, K., Watanabe, K., Wakano, K., Suzuki, N., Satomi, K. and Ishii, Y. (1983) Expansive Opendoor Laminoplasty for Cervical Spinal Stenotic Myelopathy. *Spine*, **8**, 693-699. <http://dx.doi.org/10.1097/00007632-198310000-00003>
- [5] Ozaki, M., Nakamura, M., Tsuji, O., Iwatani, A., Toyama, Y., Chiba, K., *et al.* (2013) A Rare Case of Dumbbell Meningioma of Upper Cervical Spinal Cord. *Journal of Orthopaedic Science*, **18**, 1042-1045. <http://dx.doi.org/10.1007/s00776-012-0252-6>
- [6] Hakuba, A., Komiyama, M., Tsujimoto, T., Ahn, M.S., Nishimura, S., Ohta, T., *et al.* (1984) Transuncodiscal Approach to Dumbbell Tumors of the Cervical Spinal Canal. *Journal of Neurosurgery*, **61**, 1100-1106. <http://dx.doi.org/10.3171/jns.1984.61.6.1100>
- [7] Yoshiura, T., Shrier, D.A., Pilcher, W.H. and Rubio, A. (1998) Cervical Spinal Meningioma with Unusual MR Contrast Enhancement. *American Journal of Neuroradiology*, **19**, 1040-1042.
- [8] Buchfelder, M., Nomikos, P., Paulus, W. and Rupprecht, H. (2001) Spinalthoracic Dumbbell Meningioma: A Case Report. *Spine*, **26**, 1500-1504. <http://dx.doi.org/10.1097/00007632-200107010-00026>
- [9] Chen, J.C., Tseng, S.H., Chen, Y., Tzeng, J.E. and Lin, S.M. (2005) Cervical Dumbbell Meningioma and Thoracic Dumbbell Schwannoma in a Patient with Neurofibromatosis. *Clinical Neurology and Neurosurgery*, **107**, 253-257. <http://dx.doi.org/10.1016/j.clineuro.2004.06.012>
- [10] Clarke, M.J. and Krauss, W.E. (2006) Cervical Meningioma Masquerading as Schwannoma: Two Cases and a Review of the Literature. *Journal of Neurosurgical Sciences*, **50**, 41-44.
- [11] Watanabe, M., Sasaki, D., Yamamoto, Y., Iwashina, T., Sato, M. and Motida, J. (2009) Upper Cervical Spinal Cord Tumors: Review of 13 Cases. *Journal of Orthopaedic Science*, **14**, 175-181. <http://dx.doi.org/10.1007/s00776-008-1309-4>
- [12] Leestma, J.E. (1980) Brain Tumors. *The American Journal of Pathology*, **100**, 239-316.
- [13] Simpson, D. (1957) The Recurrence of Intracranial Meningiomas after Surgical treatment. *Journal of Neurology, Neurosurgery & Psychiatry*, **20**, 22-39. <http://dx.doi.org/10.1136/jnnp.20.1.22>
- [14] Yoshihara, H., Matsuyama, Y., Goto, M., Tsuji, T., Sakai, Y., Nakamura, H., *et al.* (2003) Results of Surgical Treatment of Dumbbell-Type Neurinoma in the Cervical Region. *Rinsho Seikei Geka*, **38**, 1209-1214. (In Japanese)
- [15] Mirimanoff, R.O., Dosoretz, D.E., Linggood, R.M., Ojemann, R.G. and Martuza, R.L. (1985) Meningioma: Analysis of Recurrence and Progression Following Neurosurgical Resection. *Journal of Neurosurgery*, **62**, 18-24. <http://dx.doi.org/10.3171/jns.1985.62.1.0018>