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Case Report

Spinal Metaplastic Meningioma with Osseous Differentiation in the Ventral Thoracic Spinal Canal

Kentaro Yamane[a]*, Masato Tanaka[a], Yoshihisa Sugimoto[a], Kouichi Ichimura[b], and Toshifumi Ozaki[a]

Departments of [a]Orthopaedic Surgery, and [b]Pathology, Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, Okayama 700-8558, Japan

Ossified meningioma is classified histologically as a phenotype of metaplastic meningioma, and it is extremely rare. There are only 12 cases involving ossified spinal meningiomas in the literature. We present the case of a 61-year-old female with a primary tumor within the ventral spinal canal at T12. Although we performed a total tumor excision using an ultrasonic bone aspirator, a temporary deterioration of motor evoked potentials (MEPs) was observed during curettage with a Kerrison rongeur. The neurologic findings worsened immediately after surgery. Histologically, the tumor was diagnosed as a metaplastic meningioma with osseous differentiation. In order to avoid spinal cord injury, great care must be taken when removing an ossified meningioma located on the ventral spinal cord.

Key words: spinal metaplastic meningioma, osseous differentiation, ossified meningioma, ultrasonic bone aspirator, post-operative course

Spinal meningiomas are a common type of intradural-extramedullary tumor, accounting for 25% of all spinal cord tumors [1]. Ossified meningiomas are extremely rare and are sometimes confused with calcified meningiomas. The histological classification of ossified meningioma is a phenotype of metaplastic meningioma. Here we report the case of a patient with a metaplastic meningioma with osseous differentiation in the thoracic spine.

Case Report

A 61-year-old Japanese female presented with a 7-month history of painful muscle weakness and numbness in both lower extremities. Magnetic resonance imaging (MRI) showed an intradural tumor at the thoracic spine. She was referred to our hospital because of worsening symptoms. At the first visit to our hospital, the patient was unable to walk due to paraplegia and hypoesthesia below the level of T12. The patient's knee jerk and ankle jerk were both hyperreflexic, but she had neither bladder nor rectal disturbance.

On plain radiographs we observed an ossified tumor within the spinal canal at T12. MRI showed a low-signal-intensity lesion at this level on both T1-weighted and T2-weighted images and inhomogeneous enhancement on contrast T1-weighted images (Fig. 1). On CT scans, we could see an ovoid tumor with a high-density area located within the ventrolateral spinal canal and pressing on the spinal cord posterolaterally (Fig. 2).

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*Corresponding author. Phone:+81-86-235-7273; Fax:+81-86-223-9727
E-mail:kentaro0311@do7.enjoy.ne.jp (K. Yamane)

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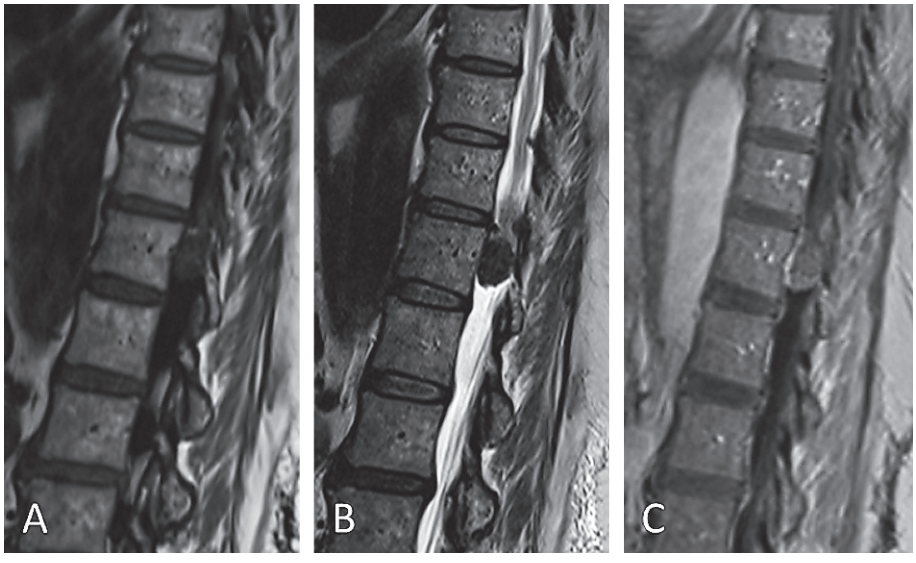

Fig. 1 T1-weighted (A) and T2-weighted (B) sagittal MR images showing homogeneous low signal intensity within the spinal canal at T12 in the patient, a 61-year-old woman. Postcontrast T1-weighted MRI (C) showing a heterogeneously enhanced tumor.

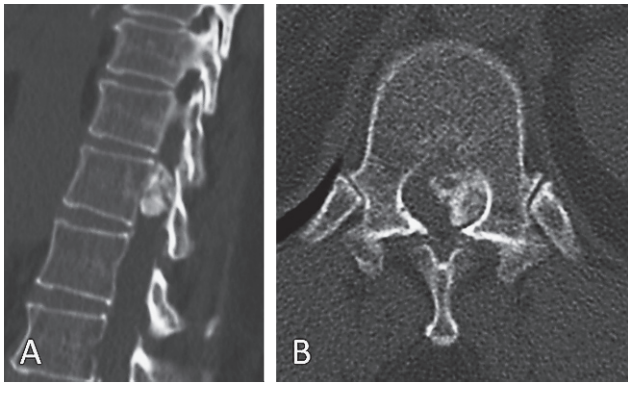

Fig. 2 Sagittal CT scan (A) showing an ovoid, high-density area within the spinal canal. An axial section (B) shows the tumor occupying more than half of the spinal canal and pressing the spinal cord laterally.

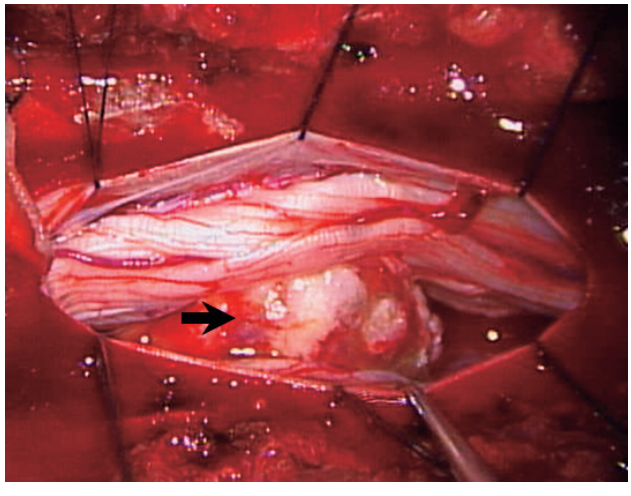

Fig. 3 Intraoperative photograph of the tumor (black arrow) at the T12 level adhered to the arachnoid membrane. The majority of the tumor was very hard.

We performed a laminectomy from the T10 to the T12 vertebrae. With the aid of a microscope, we incised the dura mater at the midline, but the arachnoid membrane adhered to the tumor. The majority of the tumor was very hard, reflecting the ossification (Fig. 3). We initially began curettage with a Kerrison rongeur, but we then observed a temporary reduction in the patient's motor evoked potentials (MEPs). Therefore, we carefully reduced the tumor using an ultrasonic bone aspirator (Sonopet®, Stryker Corp., Kalamazoo, MI, USA) and the MEP waveforms improved.

After we thinned and reduced the tumor's size, we were able to completely resect it. After their initial temporary reduction, the MEPs remained stable throughout the rest of the surgery. Although the muscle strength and dysesthesia of the patient's lower extremities worsened immediately after the surgery, her neurological condition gradually improved. At the 2-year follow-up examination, the patient could walk

with a cane and MRI revealed no recurrence of the tumor.

The histopathological examination of the tumor showed a meningotheliomatous component, which was immunohistochemically positive for epithelial membrane antigen and contained whorls and psammoma bodies. There was also a remarkable formation of cancellous bone with bone marrow (Fig. 4). The diagnosis was metaplastic meningioma with osseous differentiation (WHO Grade I).

Discussion

Ossified meningiomas are extremely rare and are distinguished from calcified meningiomas; ossification differs from calcification histologically, and calcified psammomatous bodies may not always lead to bone formation [2, 3]. According to the WHO classification, ossified meningiomas are histologically classified as a subtype of metaplastic meningioma, which is characterized by the presence of mesenchymal components.

Our review of the literature revealed only 12 cases with purely ossified spinal meningiomas (Table 1) [3–12]; we excluded reports which did not clearly differentiate between ossification and calcification. The 12 cases suggested some hypotheses as to the mechanism of ossification. The most feasible explanation is that meningioma ossification occurs secondary to metaplasia of the arachnoid tumor cells or interstitial cells rather than through psammomatous features [2, 3, 9]. Uchida *et al.* suggested that ossification

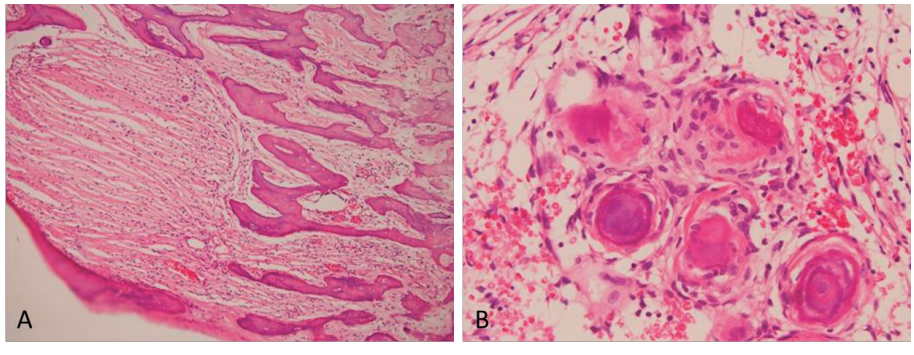

Fig. 4 Photomicrographs of a tumor section (hematoxylin and eosin). (**A**) The tumor had both meningothelial and metaplastic components (low-power field). The metaplastic subtype contained highly developed formations of cancellous bone with bone marrow. (**B**) The meningothelial component had the typical formation of whorls and psammoma bodies (high-power field).

Table 1 Data from published reports concerning ossified meningiomas

Author & Year	Age	Gender	Level	Location within spinal canal	Neurologic deterioration after surgery	Follow-up period	Tumor recurrence
Rogers, 1928	16	F	T9	Right dorsal	−	18 months	−
Freidberg, 1972	69	F	T1-T2	Ventral	+	6 weeks	ND
Niijima et al., 1993	75	F	T8-T9	Left dorsal	−	14 months	ND
Kitagawa et al., 1994	75	F	T9-T10	Right ventral	ND	ND	ND
	60	F	T7-T8	Dorsal	ND	ND	ND
Nakayama et al., 1996	45	M	C2	Left lateral	−	12 months	−
	74	F	T9	Dorsal	−	24 months	−
Huang et al., 1999	73	F	T5	Left lateral	−	18 months	ND
Naderi et al., 2001	15	M	T4	Dorsal	−	3 months	−
Liu et al., 2006	70	F	T11	Left lateral	−	24 months	−
Hirabayashi et al., 2009	82	F	L3	Left dorsal	−	60 months	+
Uchida et al., 2009	76	F	T8/T11-T12	Dorsal/left lateral	−	24 months	−
Yamane et al.	61	F	T12	Left ventral	+	24 months	−

ND: no description

may be induced by exposure to the biochemical activity of the ossification cascade or exposure to secretions from mesenchymal premature cells of osteoblast transforming factors [12].

Surgically removing an ossified meningioma can present difficulties associated with tumor adhesion and management of the dural attachment [8]. In addition, this type of tumor within the ventral or ventrolateral thoracic spinal canal is more difficult to approach and remove because of the hardness of the tumor. In 1972, Freidberg encountered a ventrally located ossified thoracic meningioma which could not be reduced in size because of its bony shell, and it could not be separated from the dura because of its firm attachment [4]. As a result of the dangerous movement of the spinal cord during surgery, a severe Brown-Séquard deficit was the post-surgical outcome. Newer and very useful surgical instruments such as ultrasonic bone aspirators simplify and accelerate the debulking of this type of tumor [13]. In our patient's case, the use of the ultrasonic bone aspirator allowed us to safely and effectively debulk the solid, ossified meningioma without stimulating the spinal cord.

Our literature review [3–12] revealed that the post-operative course of ossified meningiomas tends to be favorable. In these cases occurring within the dorsal thoracic or lumbar spinal canal, the neurologic deficits improved after surgery without major complications, and there were rare recurrences of the tumor in the short- to mid-term follow-up period (Table 1). Johnson *et al.* suggested that metaplastic meningioma grows like other grade I meningiomas with similar recurrence rates [14]. However, there was a temporary deterioration of neurological symptoms after surgery in our patient, whose tumor was located within the ventrolateral thoracic spinal canal. This was probably due to indirect spinal cord compression while we were rongeuring the hard tumor, with some resultant damage prior to our use of the ultrasonic bone aspirator. However, the patient's neurological deficits gradually improved and there is no evidence of recurrence.

Despite the progress in the design and utility of surgical instruments, the debulking and removal of spinal metaplastic meningiomas with osseous differentiation located within the ventral thoracic spinal canal are more difficult than the corresponding procedures for these tumors situated within the dorsal thoracic or lumbar spinal canal. The surgery and postoperative course of patients with a spinal metaplastic meningioma with osseous differentiation in the ventral thoracic spinal canal must therefore be conducted with particular care.

References

1. Levy WJ, Bay J and Dohn D: Spinal cord meningioma. J Neurosurg (1982) 57: 804–812.
2. Doita M, Harada T, Nishida K, Marui T, Kurosaka M and Yoshiya S: Recurrent calcified spinal meningioma detected by plain radiograph. Spine (Phila PA 1976) (2001) 26: E249–252.
3. Kitagawa M, Nakamura T, Aida T, Iwasaki Y, Abe H and Nagashima K: Clinicopathologic analysis of ossification in spinal meningioma. Noshuyo byori (1994) 11: 115–119 (in Japanese).
4. Freidberg SR: Removal of an ossified ventral thoracic meningioma. Case report. J Neurosurg (1972) 37: 728–730.
5. Hirabayashi H, Takahashi J, Kato H, Ebara S and Takahashi H: Surgical resection without dural reconstruction of a lumbar meningioma in an elderly woman. Eur Spine J (2009) 18 Suppl 2: 232–235.
6. Huang TY, Kochi M, Kuratsu J and Ushio Y: Intraspinal osteogenic meningioma: report of a case. J Formos Med Assoc (1999) 98: 218–221.
7. Liu CL, Lai PL, Jung SM and Liao CC: Thoracic ossified meningioma and osteoporotic burst fracture: treatment with combined vertebroplasty and laminectomy without instrumentation: case report. J Neurosurg Spine (2006) 4: 256–259.
8. Naderi S, Yilmaz M, Canda T and Acar U: Ossified thoracic spinal meningioma in childhood: a case report and review of the literature. Clin Neurol Neurosurg (2001) 103: 247–249.
9. Nakayama N, Isu T, Asaoka K, Harata T, Hayashi S, Aoki T, Mabuchi S and Takahashi T: Two cases of ossified spinal meningioma. No Shinkei Geka (1996) 24: 351–355 (in Japanese).
10. Niijima K, Huang YP, Malis LI and Sachdev VP: Ossified spinal meningioma en plaque. Spine (Phila PA 1976) (1993) 18: 2340–2343.
11. Rogers L: A spinal meningioma containing bone. The British journal of surgery (1928) 15: 675–677.
12. Uchida K, Nakajima H, Yayama T, Sato R, Kobayashi S, Mwaka ES, Imamura Y and Baba H: Immunohistochemical findings of multiple ossified en plaque meningiomas in the thoracic spine. J Clin Neurosci (2009) 16: 1660–1662.
13. Roux FX, Nataf F, Pinaudeau M, Borne G, Devaux B and Meder JF: Intraspinal meningiomas: review of 54 cases with discussion of poor prognosis factors and modern therapeutic management. Surg Neurol (1996) 46: 458–463; discussion 463–454.
14. Johnson MD, Stevenson CB, Thompson RC, Atkinson J and Boyer P: December 2006: 31-year-old woman with hemiparesis. Brain pathol (2007) 17: 255–257.