

Ossified thoracic spinal meningioma with hematopoiesis: A case report and review of the literature

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ABSTRACT

Meningiomas account for 25% of spinal tumors, and they are often located in the thoracic spine. The ossified subtype is even rarer, and those with hematopoiesis are rarely described. The mechanism of bone formation has not yet been clarified. A case of ossified spinal meningioma with hematopoiesis occurring in a 78-year-old woman is described. Magnetic resonance imaging revealed a lesion with a dural tail sign at the T9 level located dorsal to the spinal cord. Computerized tomography revealed a high density lesion, as high as the bone signal. Total resection was performed, and the symptoms improved. Pathological findings revealed many psammoma bodies (PBs), bone formation, and bone marrow with hematopoiesis. Both PBs and bone seemed to be based on the same background of calcified structures. This report is the second dealing with ossified spinal meningioma with hematopoiesis. The hardness of the tumor can make the operation more difficult, so that the operation should be performed carefully to avoid injuring the spinal cord.

Keywords: Spinal Cord; Meningioma; Ossification; Extramedullary Hematopoiesis; Spinal Tumor

1. INTRODUCTION

Spinal tumors are rare [1], and meningiomas account for 25% of such tumors [2-7]. Typically, meningiomas form intradural extramedullary tumors. They account for 40% of adult extramedullary lesions [2], and they are often located in the thoracic spine [2,4,6].

Ossified meningiomas account for approximately 1% of all meningiomas [8]. Even ossified spinal meningiomas are extremely rare and may be estimated to account

for 0.7% - 5.5% of all spinal meningiomas [7,9,10]. There are only a few reported cases of bone formation with hematopoiesis [11].

Most meningiomas are benign and slow growing [5,8,12,13], and the goal of treatment should be total surgical resection [14]. The ossification type is an additional issue for treatment because of difficulty in resection [6,15,16].

A case of ossified thoracic meningioma with hematopoiesis and a literature review are presented.

2. CASE REPORT

A 78-year-old woman complained of right leg pain that needed a cane for three months. Two months before the operation, pain appeared in both legs that prevented knee extension, resulting in her having to walk on her knees. Detailed examination disclosed a thoracic spinal tumor, and she was referred to our hospital for further detailed examination in September.

Thoracic magnetic resonance imaging (MRI) revealed an intradural, extramedullary tumor at the T9 level located dorsal to the spinal cord, which was severely compressed. The lesion was hypointense on both T1-weighted and T2-weighted MRI, with a dural tail sign (**Figure 1**). Computed tomography (CT) demonstrated that the lesion and the bone had the same isodensity (**Figures 2A and B**). These features suggested a spinal meningioma with areas of calcification or ossification.

At surgery, T8-10 laminectomy and excision of the outer yellow ligament were performed, and the dural sac was exposed. The dura attached to the lesion was hard and continuous with the tumor. The dura was divided into inner and outer layers, and the outer was kept.

The boundary between the tumor and the spinal cord was clear, and the tumor did not adhere to the arachnoid. Total resection of the tumor was performed with the inner dura, after which closure of the outer dura and T9-10 laminoplasty were performed.

Postoperatively, the symptoms improved, and the pa-



Figure 1. MRI (T2-weighted) of the $14 \times 9 \times 6$ mm intradural extramedullary tumor at the T9 level. It is located dorsal to the spinal cord and compresses the spinal cord from the back. The tumor shows hypointensity. The lower part of the tumor shows the dural tail sign comparable finding (arrow).

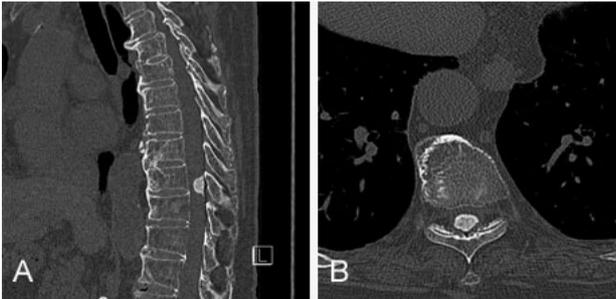


Figure 2. CT A: (sagittal section): the intradural tumor with high density, as high as the bone signal, is located dorsal to the spinal cord. There is multiple osteophyte formation and narrowing of the inner vertebral spaces; B: (transverse section): the spinal cord is compressed from behind, slight convex scoliosis is also seen on the right side, and the 5/6 right intervertebral foramen is constricted.

patient was discharged well.

3. PATHOLOGICAL EXAMINATION

The lesion measured $14.0 \times 9.0 \times 6.0$ mm. The paraffin sections, stained with hematoxylin and eosin (H & E), revealed that the tumor consisted of bland elongated cells with indistinct cell borders and short spindle nuclei. There were psammoma bodies (PBs) and bone formation (**Figure 3A**). Neither mitotic figures nor necrosis was observed. Many mature bones with a mosaic pattern and bone marrow with hematopoiesis were evident (**Figure 3B**).

There were areas with crowded PBs (**Figure 4A**), but the PBs were not always continuous with ossification. There were various combinations of patterns, with gathering of PBs with surrounding hyaline connective tissue (**Figure 4A**), mature bone entrapping a PB (**Figure 4B**), mature bone formation without a PB (**Figure 4C**), and bone formation adjacent to the PBs (**Figure 4D**). Based on these findings, the lesion was diagnosed as an ossified thoracic meningioma.

4. LITERATURE REVIEW

There have been only 16 reported cases of ossified spinal meningioma in PubMed, including the present case (**Table 1**). The age range was 15 - 78 years, with a male: female ratio of 2:14, showing clear female predominance. Six cases were reported to have bone marrow formation, but only 2 cases had evidence of hematopoiesis.

5. DISCUSSION

Intraspinal tumors are rare [1]. Meningiomas are com-

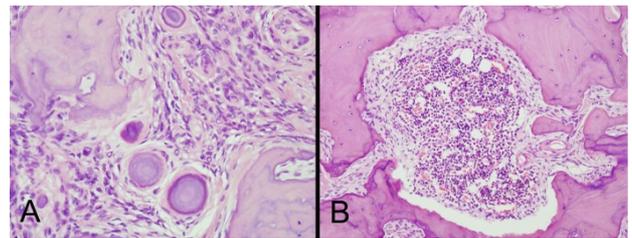


Figure 3. A: The neoplastic lesion consists of elongated cells with indistinct cell borders, short spindle nuclei, PBs, and bone formation. The lamellar bone shows mosaic pattern. (H & E stain, original magnification $\times 200$) B: Formation of mature bones including bone marrow with hematopoiesis is seen (H & E stain, original magnification $\times 100$).

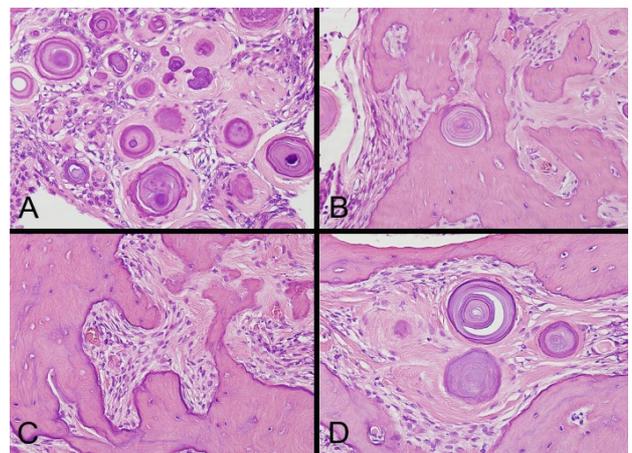


Figure 4. A: There are many PBs; some surrounded by hyaline tissue; B: Mature bone formation entrapping a PB; C: Mature bone formation without PB; D: Mature bones and PB formation (H & E stain, original magnification $\times 200$).

Table 1. Reported cases of ossified spinal meningioma.

No	Year	Age/ sex ^{#1}	Location ^{#2}	Size (mm) ^{#3}	Bone marrow ^{#3}	Symptom (Location) ^{#3}	Operation/Result ^{#4}	Outcome ^{#5}
1	1928 [17]	16/F	T9/D	15	ND	Pain (back and right side below the costal margin), dysbasia, loss of sensation, urinary incontinence	L/TR	SI
2	1972 [16]	70/F	T1-2/V	20 × 13	ND	Weakness (legs), urinary incontinence.	L(T1-4)/TR	SI
3	1993 [18]	75/F	T8-9/DL	ND	ND	Ataxic gait, progressive numbness and spastic paraplegia (lower extremities)	L(T7-10)/TR	SI
4	1994 [19]	75/F	T9-10/V	ND	+	Pain (legs), dysbasia, Numbness (legs)	SR	ND
5	1994 [19]	60/F	T6-8/D	ND	+	Dysbasia, numbness and weakness (legs)	TR	ND
6	1996 [20]	45/M	Foramen magnum, C1-3/L	ND	+	Headache (occipital), numbness (hand peripheral) hypoesthesia (arm peripheral)	L(C1-2)/TR	SI
7	1996 [20]	74/F	T9/D	ND	ND	Dysbasia, numbness (leg), hypoesthesia, urinary disturbance	TR	SI
8	1999 [9]	68/F	Cervicothoracic/ Encircling the spinal cord	ND	ND	Dysesthesias, spasms (lower extremities)	PR	SR
9	1999 [21]	73/F	T5/DL	10 × 10 × 10	+	Dysbasia, numbness and weakness (legs)	TR	SI
10	2001 [15]	15/M	T4/DL	30 × 15 × 10	ND	weakness and numbness (legs), paraplegia, hypoesthesia, urinary incontinence	L(T4, 5)/TR	SI
11	2006 [22]	70/F	T11/DL	ND	ND	Severe pain (back), progressive paraparesis, urinary incontinence	L(T10-12)/TR	SI
12	2009 [6]	40/F	T6/DL	ND	ND	Pain (back), weakness (legs), numbness (below the umbilicus) loss of sensation	L(T5-7)/TR	SI
13	2009 [22]	76/F	T8 and T11-T12/D	ND	ND	Pain and numbness (legs), weakness (legs and lower extremities), loss of sensation, urinary incontinence	TR	SI
14	2010 [11]	58/F	T6/D	15	+ and Hematopoiesis	Paraparesis	L(T5, 6)/TR	SI
15	2011 [8]	75/F	T3-T5	ND	ND	Paresthesia and hyposthenia (legs)	TR	NR
16 ^{#6}	2012	78/F	T9/D	14 × 9 × 6	+ and Hematopoiesis	Pain and numbness (legs)	L(T8-10)/TR	SI

^{#1}M: Male; F: Female; ^{#2}D: Dorsal to spinal cord; V: Ventral to spinal cord; L: Lateral to spinal cord; DL: Dorsolateral to the spinal cord; ^{#3}ND: No data; ^{#4}L: Laminectomy; R: Resection; PR: Partial resection; SR: Subtotal resection; TR: Total resection; ^{#5}SI: Symptoms improved; SR: Symptoms remained; NR: No recurrences; ND: No data; ^{#6}Present case.

mon; accounting for 25% of spinal meningiomas [2-7]. There is a difference in location depending on sex. In women, the majority of meningiomas is located in the thoracic region (80%), followed by the cervical (15%) and lumbosacral (5%) regions [2,4,6,23-25], while in men, meningiomas are located in the thoracic region (50%), followed by the cervical (40%) region [23,26].

Spinal ossified meningiomas are extremely rare, with only 16 cases in PubMed, including the present case (**Table 1**). The histogenesis of the ossification in meningiomas is not clear. There are some hypotheses about the

ossification. Barresi *et al.* presented two pathways. One was that bone formation was the final step of long-standing calcification, as frequently found in psammomatous meningiomas. The other was that the neoplastic cells surrounding bone trabeculae develop osteoblast-like properties, expressing osteopontin, as well as enchondral ossification with remodeling, by osteoclasts [8]. Uchida *et al.* mentioned that premature arachnoid cells with pluripotency, which are the origin of the neoplastic cells, differentiate into the metaplastic cells with bone formation [27].

The present case showed remarkable PBs formation. The hyalinized collagen surrounding the PBs was continuous with those attached to bone formation. There was also an area where bone entrapped a PB. Though there were no transitions between PB and bone, they seemed to be related. Both PB and bone might be based on same background tendency to develop calcified structures.

Licci *et al.* stated that the presence of lamellar bone in the lesion is sometimes reported; however, the formation of hematopoietic tissue in the marrow space is rarely mentioned [11]. Therefore, the present case is only the second case of ossified spinal meningioma with hematopoietic bone marrow. Mathew *et al.* suggested two mechanisms of de novo extramedullary hematopoiesis within meningiomas. First, the induced neoplastic growth of meningeal cells may cause erythroid differentiation in the multipotent stem cells. Second, the quiescent heterotopic cell rests that were isolated during development may develop proliferative changes and differentiation by the increased oxygen and nutritional demands of the tumor cells [28]. Licci *et al.* mentioned that a third mechanism of mesenchymal multipotentiality of meningeal or meningiomatous cells could cause hematopoietic differentiation [11].

Because of spatial restriction, symptoms are more frequent with spinal meningiomas than with cranial meningiomas [19]. The most common symptoms are pain [7, 10,12], followed by muscle weakness such as paresis or plegia, and sensory loss such as hypoesthesia, paresthesia, or anesthesia [12]. In the cases of spinal ossified meningiomas, muscle weakness is more frequent (**Table 1**).

There are some reports mentioning difficulty in resection for calcified or ossified meningiomas [6-8,15,16]. In the present case, probably because the tumor was dorsal to the spinal cord and it was not too large, the operation was not difficult.

When the tumors are resected completely, the symptoms improve [6,11,15,16,21,22,27]. However, when the tumor is not resected completely because of its hardness or large size, the symptoms remain [9].

In conclusion, spinal ossified meningiomas with hematopoietic bone marrow are extremely rare. The histogenesis of the bone and hematopoiesis is still unknown. The hardness of the tumor makes surgery more difficult, but a good outcome is expected with total resection of the tumor. Careful surgery is needed for hard tumors to avoid spinal cord injury.

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