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Case Report

Thoracic meningioma with ossification: Case report

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ABSTRACT

Background: The incidence of spinal meningiomas is 0.33/100000 population, and ossified spinal meningiomas are even less commonly encountered.

Case Description: A 64-year-old male presented with a progressive T4-level thoracic myelopathy. MR imaging revealed an intradural extramedullary mass that significantly compressed the spinal cord. The accompanying CT demonstrated hyperdensities within the lesion consistent with punctate calcification vs. ossification (i.e. consistent with histological bone formations within tumor). The patient underwent complete resection of the tumor resulting in a full recovery of neurological function within 6 postoperative weeks. The pathological specimen showed findings consistent with an ossified spinal meningioma.

Conclusion: Here, we identified a rare case of an ossified thoracic T4 meningioma occurring in a 64-year-old male.

Keywords: Myelopathy, Ossification, Spinal meningioma, Spine, Tumor

INTRODUCTION

A quarter of spinal tumors are meningiomas, and over 90% of them are benign.[1] They occur more in females (4:1 female/male ratio), and 82% are predominantly located in the thoracic spine.[20] Notably, 5% of these lesions are calcified, while <1% are ossified (i.e. exhibiting histological bone formation within the tumor).[13,17] Here, we reviewed 35 cases of ossified spinal meningiomas reported in literature and added an additional case of a T4 lesion found in a 64-year-old male who presented with a T4 lesion[1,27] [Table 1].

CASE REPORT

A 64-year-old male presented with a progressive T4-level paraparesis characterized by progressive numbness below the waistline, weakness in both lower extremities, and ataxia of gait. His neurological examination showed diffuse 4/5 bilateral lower extremity weakness with a relative T4-sensory level to pin appreciation.

Imaging

The thoracic MRI revealed a large right-sided dorsal intradural extramedullary lesion contributing to severe compression of the spinal cord at T4-level. The CT scan confirmed

Table 1: Summary of cases with ossified thoracic meningioma.

	Age/Sex	Level	Symptoms	Surgery	Clinical Outcomes	Recurrence
Roger, 1928[19]	16 F	T9	Myelopathy	GTR	Improved (3 months)	No
Freidberg, 1972[6]	69 F	T1-2	Myelopathy	GTR+dura	Improved (6 weeks)	N/A
Niijima *et al*., 1993[16]	75 F	T8-9	Myelopathy	GTR+dura	Improved (14 months)	N/A
Kitagawa *et al*., 1994[10]	75 F	T9-10	n/a	n/a	n/a	N/A
	60 F	T6-8	n/a	n/a	n/a	N/A
Nakayama *et al*., 1996[15]	74 F	T9	Myelopathy	GTR	n/a	N/A
	45 M	Upper cervical	Myelopathy	GTR	n/a	N/A
Huang *et al*., 1999[8]	73 F	T5	Myelopathy	GTR	Improved	N/A
Naderi *et al*., 2001[14]	15 M	T4	Myelopathy	GTR+dura	Improved (3 months)	N/A
Liu *et al*., 2009[12]	70 F	T11	Myelopathy	GTR	Improved (2 years)	No
Tahir *et al*., 2009[22]	40 F	T6	Myelopathy	GTR	Improved (8 months)	No
Hirabayashi *et al*., 2009[7]	82 F	L3	Cauda equina syndrome	GTR	Improved	No (5 years)
Uchida *et al*., 2009[24]	76 F	T8 and T12	Myelopathy	GTR+dura	Improved	No (2 years)
Licci *et al*., 2010[11]	58 F	T6	Myelopathy	GTR	Improved (1 year)	N/A
Chotai *et al*., 2013[3]	61 F	T4-5	Myelopathy	GTR+dura	Improved (1 month)	N/A
Ju *et al*., 2013[9]	61 F	T9-10	Myelopathy	GTR+dura	Improved (1 month)	N/A
Taneoka *et al*., 2013[23]	78 F	T9	Myelopathy	GTR+dura	Improved	N/A
Yamane *et al*., 2014[28]	61 F	T12	Myelopathy	GTR	Improved	No (2 years)
Chan *et al*., 2014[2]	64 F	T9-10	Myelopathy	GTR	Improved (6 months)	N/A
Alafaci *et al*., 2015[1]	45 M	T2-3	Myelopathy	GTR	Improved	No
	75 F	T3-4	Myelopathy	GTR	Improved	No
	86 F	T3-4	Myelopathy	GTR	Improved	No
	65 F	T7	Myelopathy	GTR	Improved	No
	72 F	C7	Myelopathy	STR	Improved	No
	40 F	T1-2	Myelopathy	STR	Improved	No
	65 F	T7-8	Myelopathy	GTR	Improved	No
	40 F	C7	Myelopathy	GTR	Improved	No
	41 F	T2-3	Myelopathy	GTR	Improved	No
Demir *et al*., 2016[5]	26 F	T9-11	Myelopathy	GTR	N/A	N/A
Cochran *et al*., 2016[4]	47 F	T8	Radiculopathy	GTR	Improved	No (22 months)
Xia and Tian 2016[26]	90 M	T10-11	Spinal cord injury after fall	GTR	N/A	N/A
Prakash *et al*., 2018[18]	60 F	T7-8	Myelopathy	GTR	Improved (6 months)	N/A
Murakami *et al*., 2019[13]	29 F	T12	Back pain, leg numbness	GTR+dura	Unchanged (12 months)	N/A
Taha *et al*., 2019[21]	22 F	T4-5	Myelopathy	GTR	Improved (6 weeks)	N/A
Wang *et al*. 2019[25]	52 F	T4	Back pain	GTR	Improved	No (2.5 years)

GTR: Gross total resection, STR: Subtotal resection, N/A: Not available

the lesion was hyperdense, consisting of intratumoral ossification [Figure 1]. The predominant differential diagnoses included ossified meningioma versus schwannoma.

Surgery

Under neuromonitoring and following a T4-T5 laminectomy, a midline durotomy was performed. This revealed an intradural

extramedullary tumor with a base adherent to the right lateral dura. The tumor was dissected off the dura allowing for gross total resection (GTR); the sensory rootlets enmeshed in the tumor capsule were easily dissected off the lesion and preserved. A watertight closure followed, and there were no intraoperative neuromonitoring changes. Within 6 postoperative months, the patient was neurologically intact except for some mild residual gait ataxia, (i.e. requiring a cane to ambulate).

Surgical pathology

Gross pathology showed the lesion was irregular, tan, and rubbery, measuring 9 × 10 × 13 mm. On microscopy, there were meningothelial cells with oval to spindle-shaped nuclei containing occasional intranuclear pseudoinclusions. Frequent swirls of psammoma bodies were also seen. Additional areas showed more extensive "ossification" (i.e. bone formation, osseous metaplasia). As the tumor showed little mitotic activity, and there were no areas of hypercellularity, the final diagnosis was for a WHO grade I (benign) meningioma [Figure 2].

DISCUSSION

One percent of spinal meningiomas are ossified. The majority occur in females[1,20] [Table 1]. Ossification of spinal meningiomas is attributed to metaplasia of the arachnoid.[21] Estrogen deficiency is also suspected to intensify the calcification/ossification of meningiomas where there are necrotic fibroblasts and an elevated number of collagen fibrils.[21]

Incidence and prognosis for ossified spinal meningiomas

Of the 35 ossified meningiomas identified in the literature, only four occurred in males[21] [Table 1]. GTRs best correlate with marked neurological improvements.[27] The resection of the involved adjacent dura warrants duroplasty, which increases the complexity of the procedure.[1,21,27] Perhaps, a complete ossification, firmness of the tumor, and size can increase surgical morbidity because of difficulty to manipulate the tumor without compressing the spinal cord. In some cases, GTR is unsafe because of an absent distinct plane between the tumor and the spinal cord.

Although most cases reported favorable outcomes, others reported major perioperative morbidities, including paraplegia, complete sensory loss, cerebrospinal fluid leakage, and stroke. Despite these complications, following appropriate treatment/medication, many patients sustained adequate recoveries.[21] However, most patients did not fully recover function and exhibited various degrees of neurological symptoms.

Symptom onset of ossified spinal meningiomas

Most of the 35 cases of ossified meningiomas presented with progressive myelopathy that worsened over a prolonged period. The clinical presentation was nonspecific and slow-progressing,

Figure 1 : Pre-operative imaging. (a) sagittal MRI contrast-enhancing intradural extramedullary lesion at T4 with severe compression of the spinal cord, (b) sagittal CT demonstrates hyperdense signal within the tumor suggesting calcifications.

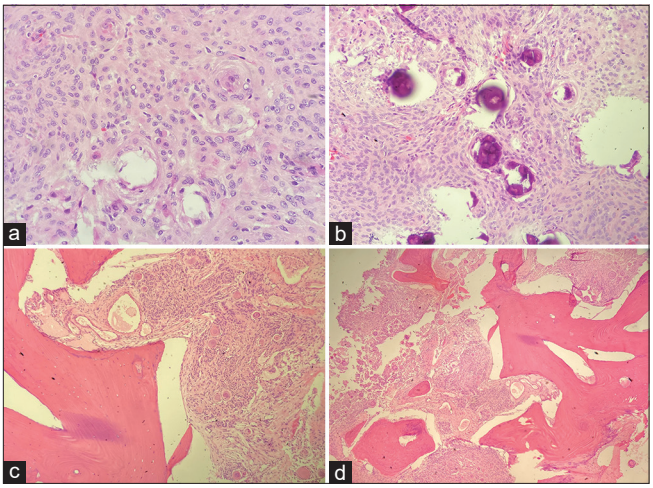

Figure 2: Hematoxylin and eosin (×20) images demonstrate (a) meningothelial cells with swirls, (b) numerous psammoma bodies, (c and d) ossifications.

therefore raise patients' concern only when severely symptomatic. MRI identifies the size/extent of the mass, and CT studies are utilized to identify small calcifications/ossification. However, imaging modalities are unable to differentiate between ossification and calcification. The final diagnosis is made based on histopathologic evaluation.

Prediction of local recurrence

Only 17 cases had reported long-term follow up, and none of the patients had a recurrence. Interestingly, there was no recurrence in patients with subtotal resection [Table 1].

CONCLUSION

Only 1% of spinal meningiomas are ossified, and few occur in males. Here we present a 64-year-old male with a T4

ossified meningomas responsible for a thoracic paraparesis that resolved following gross total tumor resection.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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