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Case Report

Dorsal extradural meningioma: Case report and literature review

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Abstract

Background: Extradural spinal mass lesions are most commonly metastatic tumors. Extradural meningiomas are rare, accounting for approximately 2.5–3.5% of spinal meningiomas; intraoperatively, they are easily mistaken for malignant tumors, especially in the *en plaque* variety, resulting in inadequate surgical treatment.

Case Description: Our case is one of the first to describe a patient with two purely extradural meningiomas, one each between D3–D4 and between D5–D6 vertebral levels. Surgical resection was radical, and pathologically both lesions were meningothelialmeningiomas.

Conclusions: Reviewing the literature, we discuss the pathogenesis, treatment strategies, and long-term behavior of these uncommon lesions.

Key Words: Extradural extramedullary neoplasms, extradural meningioma, extradural spinal tumors

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INTRODUCTION

Vertebromedullary tumors are classified as either extradural or intradural. Intradural tumors are further divided into intramedullary or extramedullary. The most common intradural extramedullary neoplasms are schwannomas, neurofibromas, and meningiomas. Extradural tumors are most commonly metastatic lesions.[38]

Meningiomas account for 25–46% of primary spinal tumors,[14] with a peak age of 40–70 years, a female ratio = 4:1 overall, and the most frequent location in the thoracic spine.[24] Extradural spinal meningiomas are very rare and account for only 2.5–3.5% of all spinal meningiomas.[40,51,66] Multiple spinal meningiomas also occur rarely.

We report a rare case of two purely extradural thoracic spine meningiomas in a young woman suffering from meningiomatosis. We discuss the features and clinical management of entirely extradural meningiomas.

CASE REPORT

A 39-year-old woman, suffering from meningiomatosis, was operated in our institute several times. The first operation was performed in 2006, when she underwent exeresis of a paramedian meningioma (meningothelialmeningioma) arising in the right paramedian region. In 2009, she presented with a gran mal seizure and left hemiparesis; magnetic resonance imaging (MRI) revealed multicentric meningiomas in the right parietal paramedian and bilateral falcine

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regions. These lesions were partially removed and the postoperative course was good, with an improvement in her seizures and weakness. In addition, in this case, the histological diagnosis was meningothelialmeningioma. Therefore, after operation, the patient was subjected to radiotherapy and a close neuroradiologic follow-up. Since February 2015, she noted progressive numbness and weakness in both lower extremities with gait disturbance. She denied sphincter disturbance.

Spine magnetic resonance imaging (MRI) showed an extramedullary mass, located between D3–D4 vertebral levels, extending to the left D3–D4 foramen [Figure 1]. Another extramedullary mass was located between D5–D6 vertebral levels, extending to the right D5–D6 foramen [Figure 2]. Both lesions were hypointense to the spinal cord on T1-weighted images, T2 hyperintense, displayed important enhancement after Gadolinium injection, and compressed the spinal cord dorsally.

Neurological examination revealed a severe paraparesis with motor strength of 2/5 in both legs (left more than right) and a T6 sensory level bilaterally. A mild weakness in her arm was also noted (outcome of previous intervention). The following deep tendon reflexes were exaggerated: Bilateral knee jerk, bilateral ankle jerk, and bilateral medial hamstrings. Left-sided Babinski reflex was noted. General physical examination was normal.

Operation

Using an operative microscope, the patient underwent a radical surgical excision of both lesions by D4 laminectomy and D5 left partial hemilaminectomy. Two purely extradural tumors were found intraoperatively; lesions were found to be gray, with hard consistency, along with calcifications and signs of bone erosion. Intraoperative findings were suggestive of a metastatic extradural and vertebral secondary location (even if no primary lesion was found in the preoperative laboratory and radiological evaluation). Extradural masses were completely removed and dural basements were cauterized [Figures 3 and 4]. Pathological examination of masses revealed, in both cases, a meningothelialmeningioma [Figure 5]. Ki67 was expressed in 2% of neoplastic cells.

Postoperatively, there was a marked improvement in the muscle strength of the lower extremities; follow-up neuroimaging revealed complete removal of lesions and no spinal instability. No apparent tumor recurrence or regrowth was detected in a follow-up study 6 months later.

DISCUSSION

Exclusively extradural meningiomas are very rare, accounting for 2.5–3.5% of spinal meningiomas.[40,45,51,66] Reviewing the literature, 44 studies have described 100 patients with extradural spinal meningioma [Table 1].

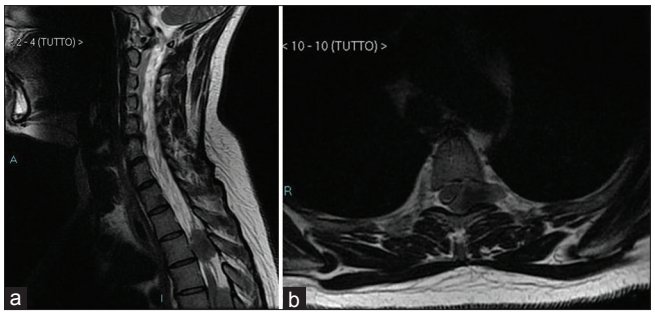

Figure 1: (a) Sagittal magnetic resonance (MR) image showing an enhancing extramedullary mass located between D3–D4 vertebral levels; (b) axial MR image showing an enhancing extramedullary mass with spinal cord displacement and compression, and extending to the left D3–D4 foramen

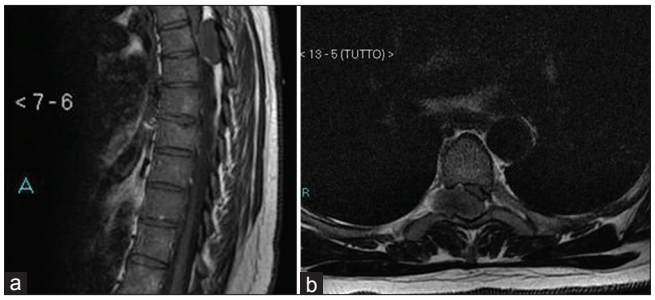

Figure 2: (a) Sagittal magnetic resonance (MR) image showing an enhancing extramedullary mass located between D5–D6 vertebral levels; (b) axial MR image showing an enhancing extramedullary mass with spinal cord displacement and compression, and extending to the right D5-D6 foramen

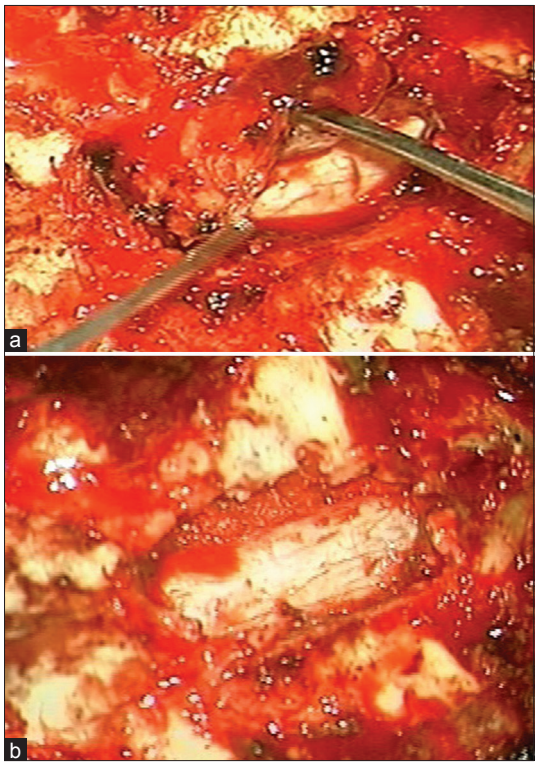

Figure 3: After D4 laminectomy, the mass was removed (a); the lesion was completely extradural (a); the underlying dura mater was intact (b)

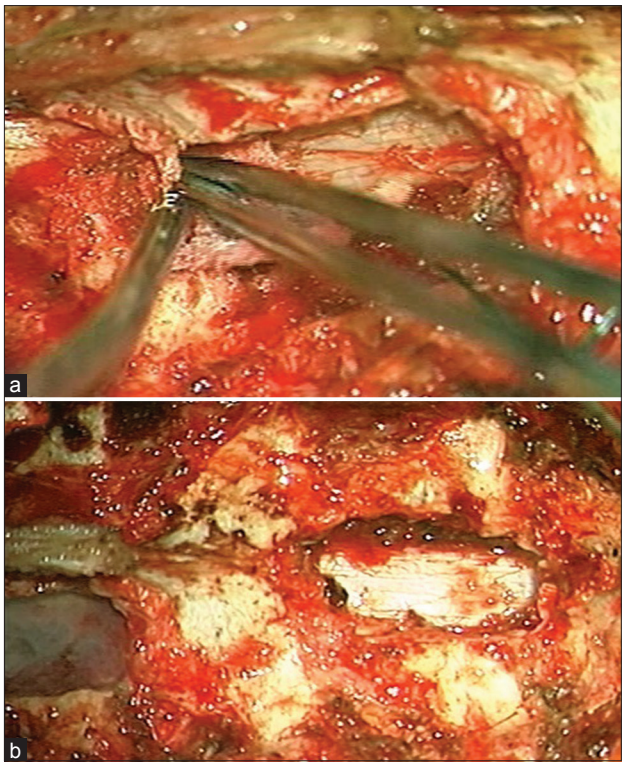

Figure 4: After D5 left partial hemilaminectomy, the mass was removed, it was completely extradural (a); the surgical field at the end of meningiomas exeresis: the dura mater was comletely intact (b)

However, because many reports used different criteria to differentiate between partially and completely extradural lesions, it is difficult to get their real frequency, which may be lower than that already presented.[45,64] In our literature review, patients with extradural meningiomas, ranged in age from 8 to 76 years old (mean, 41 years); the majority of patients were female (60%), and the lesions occurred most commonly in the thoracic spine.[9,10,22,35,36,58] These characteristics, as well as histology and clinical behavior, seem to be no different from their intradural counterparts.[45,52]

Several theories have been postulated for the pathogenesis of purely extradural meningiomas. They are believed to arise from ectopic arachnoid cells around the periradicular nerve root sleeve, where the spinal meninx merges directly into the dura,[55,68] as seen with other extracranial meningiomas such as the nose or skin.[20,54] Other authors have postulated that the periradicular dura, being less thick, may contain vestigial remnants of the superficial layers of the embryonal arachnoid mater and villi, explaining the extradural location and root proximity of some meningiomas.[55,64] It has also been suggested that island of arachnoidal tissue might migrate into the extradural space, as seen with intraorbital meningiomas that have no association to the sheath of the optic nerve.[20,54,55,64]

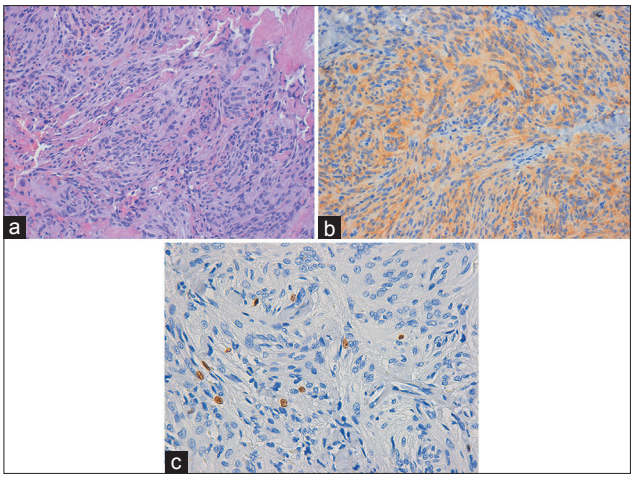

Figure 5: Histology. Hematoxylin and eosin staining (a) shows the histologic pattern of meningothelial meningiomas and polygonal cells with ill-defined cytoplasm, immunohistochemically EMA+ (b), with low Ki67 proliferation index (c)

Concerning neuroimaging, MRI is the technique of choice for the diagnosis of spinal meningiomas; it clearly defines the mass and its relation to the spinal cord.[37,65] Mostly, the lesion appears iso or hypointense on T1 MRI images, hyperintense on T2 sequences, with homogeneous enhancement after Gadolinium injection. Klekamp and Samii sustained that MRI imaging led to earlier diagnosis of spinal meningiomas by 6 months.[29] Prior to the advent of MRI imaging, mielography was the best imaging technique for the diagnosis of spinal meningiomas.[23,30] Bone window computed tomography (CT) and/or Radiography may show calcifications, bone destruction or erosion, increase in interpedicular distance, etc.[64] These radiographic characteristics, associated with extradural location, gross intraoperative appearance, and rapid progression of symptoms may induce many investigators to confuse extradural meningiomas with extradural spinal metastases, especially in the case of the *en plaque* variety.[64] In our case, we performed a gross-total resection, however, this is not always possible and the erroneous idea of a metastatic/malignant lesion may alter surgical approach, leading some surgeons to perform two surgical operations; at first a palliativesurgery for a presumed metastatic lesion, and later, a second operation for a complete resection of the tumor.[64] Therefore, intraoperative histology is mandatory for choice of the treatment strategy; if the intraoperative diagnosis is unequivocal for meningioma, the surgeon should consider gross total exeresis of the tumor. Based on the pathogenetic hypothesis that extradural meningiomas arise from the dural sleeve and not from the external surface of the dura, these tumors can be stripped-off from the spinal dura, without the need to excise the dura, as demonstrated by Savardekar.[55]

Table 1: Review of cases of epidural meningiomas described in literature

Article Author	Epidural with an intradural component/total	Exclusively epidural	Age	Gender	Tumor location	Bony changes on Neuroimaging
Tissier, 1898[62]	1		14	M	C2-C5	
Soderbergh and Sundberg, 1916[56]	1		59	M	C3-C4	
Mass, 1918[36]	1		54	F	D4-D6	
Naffziger and Ha, 1933[42]	4		54	M	D8	
			60	F	D9	
			55	M	C6-D2	
			34	F	C7-D2	
Enderle, 1934[18]	1		44	M	L3-L4	
Noisi, 1936[43]	1					
Ingraham, 1938[27]	1		10	M	C3-C5	
Rasmussen, 1940[48]	10/140					
Elsberg, 1941[17]	4/73					
Oddsson, 1947[45]	1		32	M	L/S	
Bull, 1953[9]	6/59					Pedicle destruction (1 case)
Henschen, 1955[26]	3		42	F	C5	
			17	F	D1	
			31	M	D5	
Arseni, 1958[3]	9/114					
Rand, 1960[47]	1	1	8	F	dorsal	Destruction of left D6-D7 hemilamina and erosion of pedicle
Lombardi, 1961[35]	3/71	3				Widened intervertebral foramen
Haft, 1963[25]	1	1	24	M	D5-D7	Normal
Early, 1966[16]	1	1	14	M	D3-D5	Erosion of D4 body
Soo, 1966[58]	2	1	64	F	D4-D6	Normal
		0	7	M	C1-D7	Widened pedicles
Vakili, 1967[64]	1		61	M	D1	
Pecker, 1967[46]	3		25	F	C2-C5	Normal
			43	F	C6-D1	Enlargement of C7-D1 foramen
			65	F	D4-D6	Erosion of D3-D5 pedicles
Rath, 1967[49]	1	1	20	F	C3-C6	Normal
Abbott, 1968[1]	1	0	57	M	D4	
Hallpike, 1968[24]	1	1	33	M	D6-D7	Erosion of the right D6 pedicle
Singh, 1968[55]	1		35	F	D4	
Fortuna, 1969[19]	4	1	53	F	D7-D10	
		1	74	F	D7-D8	
		1	40	F	D4-D6	
		1	31	M	D1-D2	
Mittal, 1960[40]	1		40	F	D12	
Balaparameswararao, 1970[5]	3/14		40	F	L3-L5	
			54	F	L4-L5	
			30	F	D3-D4	
Calogero, 1972[10]	4	0	40	F	D4	
		0	54	M	L3	
		0	28	F	D9-D11	
		0	33	M	D7	
Borghi, 1973[7]	5		28	F	C7-D2	Erosion of C7 and D1 pedicles
		0	23	F	D1	Normal
			32	F	C5-D1	Normal
		0	28	F	D5-D6	D6 pedicle thinned

Contd...

Table 1: Contd...

Article Author	Epidural with an intradural component/total	Exclusively epidural	Age	Gender	Tumor location	Bony changes on Neuroimaging
			60	F	D3-D4	Erosion of pedicle
Bret, 1976[8]	2/60					
Sartor, 1977[52]	1	1	29	M	C1-C4	
Roux, 1996[50]	7/54	5				
King, 1998[31]	4/78	2				
Kumar, 1980[32]	1	1	55	M	D6	
Motomochi, 1980[41]	1		14	M	C4-C7	
Stern, 1980[60]	3	1	49	F	D3-D6	
		0	60	M	D2-D6	
			56	F	D2-D3	
Kaya, 1982[28]	1		11	M	C4-C7	
Levy, 1982[34]	7/97	0				In 2 cases, vertebral posterior arc and pedicle disruption
Milz, 1983[39]	2	1	70	F	D4-D6	Normal
		1	45	F	D8	Normal
Kyoushima, 1987[33]	1					
Stechison, 1987[59]	2	0	76	F	D5-D6	Calificarachnoiditis
			63	F	D12-L1	-
Solero, 1989[57]	9/174	9				
Chen, 1992[11]	1		14	F	C2-C6	
Di Rocco, 1994[15]	1		14	F	D6-D7	
Christopherson, 1997[12]	1		13	F	D2-D3	
Sato, 1997[53]	1	1	39	M	C1-C3	
Yoshiura, 1998[67]	1	0	16	F	C2-C4	
Achari, 2000[2]	1					
Gamache, 2001[21]	1					
Messori, 2002[38]	1	1	14	F	C5-C7	Calcifications
Zevgaridis, 2002[68]	1	1	75	F	D11-D12	
Cohen-Gadol, 2003[13]	7/40	0				
Takeuchi, 2006[61]	1	1	50	M	C1-C4	Enlargement of the C3-C4 foramen
Yamada, 2007[66]	1	1	22	F	C1-C5	Calcification in the spinal canal; tumor infiltration over both the sides of the transvers processes
Barbanera, 2007[6]	1	0	53	F	C7-D1	C6-C7 vertebral bony destruction
Frank, 2007[20]	1	1	45	F	C5-C7	Enlargement of C6-C7 foramen
Santiago, 2009[51]	1	1	42	M	D2-D3	Bone remodelling of the left posterior segment of the D3 body
Tuli, 2012[63]	1	1	42	F	D4-D6	Normal
Savardekar, 2014[54]	2	2	35	F	C3-C6	C4 and C5 lateral spinous processes infiltration
			23	F	D4-D5	Normal
Nsir, 2014[44]	1	1	70	M	D5	T5 body signals changes

M: Male, F: Female. Blank cells in column 3 indicate that we had no information if the lesion was only epidural or intra and epidural; Blank cells in column 7 indicate that there was no mention of bony changes in the text

Various studies have provided contradicting reports for the long-term prognosis of patients with extradural spinal meningiomas. Some authors have asserted the aggressive behavior of these tumors, others have demonstrated these meningiomas to be benign.[64] However, we suggest that their behavior is related to their pathological World Health Organization (WHO) grade, and a bad prognosis may be caused by a difficulty in a gross-total resection of the tumor because of its bony involvement and/or paraspinal extension,[55] as well as by the genetic predisposition to form neoplasms. A complete or partial loss of chromosome 22 was noted in more than 50% of patients with spinal meningiomas.[4,23,31] Arslantas sustained a relationship between some abnormalities of cancer-related genes located on 1p, 9p, 10q, and 17q and the etiology of spinal meningiomas.[4]

CONCLUSION

Purely extradural meningiomas are very infrequent but they should be included in the differential diagnosis of extradural lesions. They are easily mistaken pre and intraoperatively for metastatic tumors, with possible consequences on the proposed surgical treatment, and therefore, on the morbidity. Because prognosis is related to the extent of resection, we believe it would be better to completely remove the lesion, if considered safe. In case of doubts, an intraoperative pathology support could be useful. Finally, because of opposing views in the long-term prognosis and rate of surgical cure, it is very important to be aware of these lesions, undergoing patients to very close follow-ups.

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Conflicts of interest
There are no conflicts of interest.

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