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Spinal Clear Cell Meningioma without Dural Attachment: Case Report and Review of Literature

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Abstract

Background: Clear Cell Meningioma (CCM) is a very rare histologic subtype of meningioma usually affecting younger patient. The reported data on spinal CCMs are extremely rare. Furthermore, only few cases without dural attachment have been reported in literature.

Methods: We report a case of a 58-year-old female patient who was presented to our department with pain in her lower back and bilateral sciatica for 6 months. Magnetic resonance imaging (MRI) showed an intra-dural well demarcated lesion at L3. The mass had an iso-intense signal on both T1 and T2 weighted images with intense and homogeneous gadolinium enhancement.

Result: Via a posterior approach, a total resection was possi¬ble due to lack of dural adhesion of the tumor. Histologic diagnosis was clear cell meningioma. Patient's recovery course after the operation was uneventful.

Conclusion: Gross total resection is recommended in treating spinal CCM whenever possible. However, radiotherapy could be considered for patients who have undergone STR or younger patients, regardless of the extent of resection.

Keywords: Clear cell; Meningioma; Spine; Dural attachment; Lumbar

Introduction

Clear Cell Meningioma (CCM) is a rare histologic subtype of meningioma. It accounts for less than 1% of all meningiomas [1]. Compared to ordinary meningiomas, CCMs have higher recurrence rate (~50%) and higher tendency to metastasize (4.1%) [2,3]. For these reasons, the World Health Organization (WHO) classification system has classified them as grade II tumors. Among the few CCMs' reported cases, the majority of them had an intracranial location. Intra-spinal CCMs were even rarer since only less than 100 cases have been reported until 2019 [4]. Most meningiomas are attached to the dura mater, therefore it is exceptional to witness a meningioma without any dural attachment. To the day we speak, only 19 cases of non-dura attached spinal CCM have been reported [4]. In this article, we present an unusual case of a spinal CCM characterized by the absence of dural attachment.

Case Report

A 58-year-old female patient was admitted with complaints of low back pain and bilateral sciatica. Her symptoms started 6 months before her admission and have worsened over time. The patient took a symptomatic treatment without any amelioration. On examination, her general condition as well as her cardiorespiratory system was normal. The neurologic examination did not reveal any specific signs except a classical lumbar spinal syndrome. Nevertheless, it is important to mention that there was no neurologic deficit. A provisional clinical diagnosis of lumber disc herniation was made and a Magnetic Resonance Imaging (MRI) was suggested. The MRI showed an intra-dural lesion at L3 measuring 31 mm of height. The mass was well demarcated and had an isointense signal on both T1-and T2-weighted images with intense and homogeneous gadolinium enhancement (Figure1). There was no foraminal extension and no associated bone destruction.

Based on the clinical condition of the patient and the MRI findings, we decided to operate the patient. We started with a L2-L3 laminectomy. The intra-dural exposure revealed an elliptic, encapsulated yellow mass of firm consistency. The tumor was found

to be draped by and adhered to the nerve roots without any dural attachment. After an easy dissection, we managed to perform a total resection. On microscopic examination (Figure 2), the tumor was composed of layers of polygonal cells with a clear glycogen-rich cytoplasm and monomorphic non-mitotic nuclei. There were prominent perivascular and interstitial collagen. Whorl formation and psammoma bodies were absent. The pathological diagnosis was a CCM (WHO II). The patient's postoperative course was uneventful. All of her symptoms did disappear and she was able to get up the first day after the operation. A postoperative MRI was scheduled but due to the exceptional situation of COVID19 pandemic, the patient was given an appointment in late 2020 as her case wasn't judged as urgent.

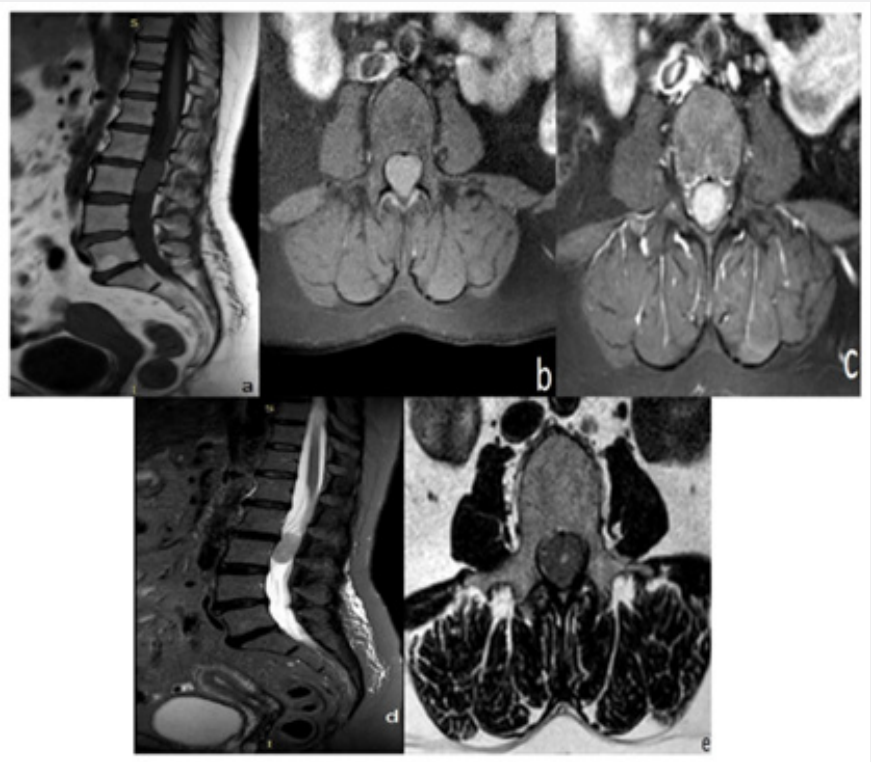

Figure1: (a,b)Intra-dural mass at L3 isointense on T1 on sagittal and axial section (c) and after injection of gadolinium (d,e)and hyperintense on T2 on sagittal and axial section.

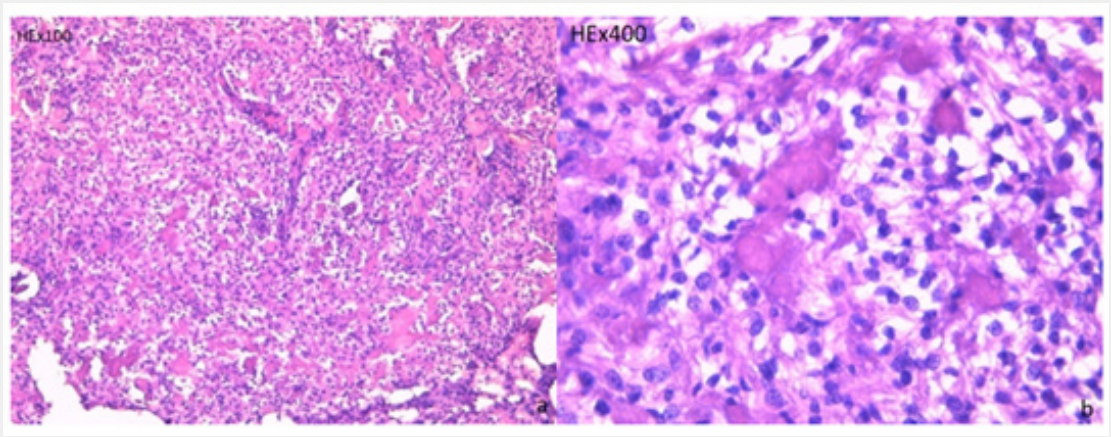

Figure 2: Pathological features of spinal clear cell meningioma: Tumor cells in classical CCM area with distinct cellular outlines and abundant clear cytoplasm with no mitosis and no necrosis area (a: H&E staining with original magnifications x 100; b: H&E staining with x400).

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Discussion

Table1: Information summary of the reported intra-spinal CCMs in English literatures.

Patient no.	Reference	Report year	Age (years)	Sex	Location	Dural Attachment	Surgical Treatment	Adjuvant Therapy	Time of Recurrence (Months)
1	Present case	2020	58	F	L3	NO	GTR	None	No
2	Wu et al. (23)	2019	7	F	T11-L1	YES	GTR	None	No
3			7	F	L2-L4	YES	GTR	None	No
4			4	M	T11-T12	YES	GTR	None	No
5			50	F	T11	YES	GTR	None	No
6			23	M	L5-S3	YES	GTR	None	No
7			52	F	C5	YES	GTR	None	No
8			43	F	L3-S3	YES	STR	None	No
9			21	M	L5	YES	GTR	None	No
10			20	F	L4-L5	YES	GTR	None	No
11			16	F	T12-L1	YES	GTR	None	No
12	Li et al. (22)	2019	50	M	C3-T1	YES	GTR	None	No
13			28	M	T10	YES	GTR	None	No
14			20	F	L3	YES	GTR	None	No
15			15	F	L3	YES	GTR	None	No
16			34	F	L4	YES	GTR	None	No
17			37	F	L5-S3	YES	GTR	None	No
18			16	M	L5-S1	YES	GTR	None	No
19			53	M	L4-S1	YES	GTR	None	No
20			27	F	C1-C2	YES	STR	None	120
21			16	F	T12-L1	YES	STR	None	120
22			14	F	L4	YES	STR	None	24
23			35	M	L3-S2	YES	GTR	None	12
24		2018	50	M	L5-S3	YES	GTR	None	18
25	Yang et al. (19)		56	M	L1-S2	YES	GTR	None	40
26	Inoue et al. (24)	2018	5	M	L5-S1	No	GTR	None	No
27	Kawasaki et al. (25)	2018	8	F	L2	No	GTR	None	No
28	Tausiede-Espariat et al. (26)	2018	13	F	C4-C5	NA	GTR	None	No
29			26	F	Lumbar	NA	GTR	None	No
30	Wu et al. (27)	2017	14	M	C1-C2	YES	GTR	None	No
31			7	F	L2-L4	YES	GTR	None	No
32			16	F	T6-T7	YES	GTR	None	No
33	Smith et al. (28)	2017	22	M	L4-S2	NA	GTR	None	No
34			33	F	L5-S2	NA	GTR	None	83
35			48	M	L5-S2	NA	GTR	None	No
36			19	F	T12	NA	GTR	None	No

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37			10	F	L1-L2	NA	GTR	None	No
38	Kim et al. (29)	2016	57	F	Thoracic	YES	GTR	None	No
39	Li et al. (1)	2016	21	M	L5	YES	GTR	None	No
40			43	F	L3-S3	YES	STR	None	No
41			7	F	T11-L1	YES	GTR	None	No
42			7	F	L2-L4	NO	GTR	None	No
43			4	M	T11-T12	YES	GTR	None	No
44			20	F	L4-L5	YES	GTR	None	No
45	Evans et al. (30)	2015	3	M	L1-L2	NA	GTR	None	No
46	Meguins et al. (31)	2014	17	F	L5-S1	NA	GTR	None	No
47	Wang et al. (17)	2014	35	M	L5-S1	NA	GTR	RT	No
48	Balogun et al. (32)	2013	3	M	L2-L5	NA	GTR	None	9
49	Zhang et al. (33)	2013	26	F	Temporal lobe + T12-L1	NO	GTR	None	No
50	Kobayashi et al. (34)	2013	43	M	L2-L3	NO	GTR	None	No
51	Ko et al. (35)	2011	34	F	L2-L3	NO	GTR	None	No
52	Prayson et al. (36)	2010	42	M	Cervical	NA	GTR	None	No
53	Tong-tong et al. (37)	2010	35	F	C7	YES	GTR	None	No
54	Nakajima et al. (38)	2009	21	F	L2-L4	YES	GTR	None	No
55	Colen et al. (39)	2009	13	F	L4-L5	NA	GTR	RT	No
56	Greene et al. (40)	2008	3	F	Cervical	NA	GTR	None	No
57	Jain et al. (41)	2007	26	F	Cauda equina	YES	GTR	None	No
58	Vural et al. (42)	2007	4	F	C1-C2	NA	GTR	None	No
59	Park et al. (10)	2005	65	F	T9-T10	NO	GTR	None	No
60	Dhall et al.(16)	2005	32	F	Thorac-ic-lumbar	NA	GTR	None	No
61	Epstein et al. (43)	2005	41	F	L3-L4	NO	GTR	None	No
62	Jia et al. (44)	2005	40	F	L1-L2	NA	GTR	None	No
63	Oviedo et al.(15)	2005	7	M	L2-L3	NO	GTR	None	No
64	Liu et al. (20)	2005	2.2	M	T10-L1	YES	GTR	None	60
65	Chen et al. (18)	2004	41	F	L4-L5	NO	GTR	None	No
66	Boet et al. (45)	2004	34	M	L4-S3	YES	STR	RT	No
67	Payano et al. (14)	2004	24	M	L3-L4	NO	GTR	None	No
68			19	F	L3	NO	GTR	None	No
69	Carrà et al. (46)	2003	1.8	M	T11-L4	NO	GTR	None	60

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70	Yu et al. (47)	2002	1.2	F	T12-L2	YES	GTR	None	8
71	Jallo et al. (2)	2001	8	F	L3-L4	NO	GTR	None	6
72			1.8	F	C3-C5	NO	STR	None	2.3
73	Park et al. (48)	2000	1.2	F	T12-L2	NA	GTR	None	8
74	Heth et al. (49)	2000	7	F	L4-L5	NA	GTR	None	No
75	Alameda et al. (50)	1999	42	M	Lumbosa-cral region	YES	GTR	None	No
76	Dubois et al. (51)	1998	10	F	L1-L4	NO	GTR	None	6
77	Matsui et al. (52)	1998	9	F	L2	NO	GTR	None	4
78	Cancès et al. (53)	1998	9	F	L1-L4	YES	GTR	RT	5
79	Pimentel et al. (54)	1998	55	M	Cervical	NA	GTR	None	No
80			21	F	Lumbar	NA	GTR	None	No
81	Holtzman et al. (55)	1996	32	M	L3-L4	NO	GTR	None	No
82	Prinz et al. (56)	1996	38	M	Sacro-coc-cygeal	NA	GTR	None	4
83		1996	23	F	L5	YES	GTR	None	No
84	Zorludemir et al. (6)		36	F	L2-L5	NA	GTR	None	No
85			17	F	L4-L5	NO	GTR	None	No
86			34	M	L4-S1	YES	GTR	None	12
87			9	F	L3-L5	YES	GTR	None	6
88			47	M	L3-L4	YES	GTR	None	36

F: Female, M: Male, NA: Not Available, GTR: Gross Total Resection, STR: Subtotal Resection, RT: Radiotherapy.

First reported by Manivel and Sung [5] in 1990, CCM is one of the rarest histologic subtype of meningiomas, representing 0.2-0.8% [6-8]. Zorludemir et al. (6) & Oviedo et al. [7] found that this subtype of meningiomas had a higher local recurrence rate and a more aggressive clinical course compared to ordinary meningiomas. These differences led to the classification of a grade II type tumor in 2016 [9]. Around 300 cases of CCMs are reported in the literature, most of them had an intracranial location. The reported data on intraspinal CCMs are extremely rare. To date, only 87 cases of spinal CCMs have been reported since 1996, and only 19 of them had no dural attachment. Detailed information is documented in (Table 1). However, it is noteworthy that the proportion of spinal to total meningiomas is higher for CCMs (45%) than for other meningiomas (the highest being 20% of the reported cases) [10]. In 2019, Zhang et al. (4) found that CCMs mostly attack young patient with a mean age at resection of 24 years old. Amazingly, 36 (42.9%) patients suffered from spinal CCM at an age of under 18 years. As it is the case of other meningiomas, CCMs have a slight female predominance with a female to male ratio at 1.7:1 (53 vs 31). In our review, 37 (42%) patients were under 18 years old and the mean age at resection was 24.6 years. 56 (63.6%) patients were female, 32 (36.4%) patients were male and the female-to-male ratio was 1.75:1. As for the most affected spinal region, the ordinary meningiomas were located in the thoracic region [11-13] whereas (66.7%) of CCMs were located in the lumbar region [4]. The imaging features of CCMs is very similar to those of ordinary meningiomas [15,16]. The MRI of CCMs often reveals an intradural-extramedullary, well-demarcated, homogeneously enhanced mass. It is isointense on both T1-and T2-weighted images, and demonstrates fairly homogeneous enhancement after the injection of gadolinium [8, 17]. Some cases showed foraminal extension and lacking of dural at-tachment, just like schwannomas [18] and other cases showed even an intramedullary CCM [10]. Because of radiologic and gross morphologic similarities between the spinal meningiomas and other intradural extramedullary spinal tumors such as schwannomas, neurofibromas, and ependymomas, an accurate histological diagnosis is mandatory. Just like any other intra-spinal lesion, the bone destruction by a CCM is rare, it was reported only two times until 2018. Both cases were reported by Jian Yang et al. [19]and they showed serious bone destruction at admission. The rarity of bone destruction may be explained with the fact that neurological symptoms usually appear before bone involvement due to the limited space of spine canal, especially in the lumbosacral

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region. Histologically, CCM contains layers of clear, glycogen-rich (Periodic acid-Schiff positive, diastase-labile), polygonal cells forming only a few vague whorls [6]. Its abundant glycogen is the reason why it is called a clear cell meningioma [14]. These cells are almost Immunoreactive (IR) to Epithelial Membrane Antigen (EMA) and vimentin, while not IR to GFAP, S-100, CK and SMA, with a ki67 index varying from 0 to 40%. The over expression of EGFR, PDGF-receptor and VEGF in CCMs promotes meningioma cell proliferation, a key process in meningioma angiogenesis, the formation of peritumoral edema, and the tumor aggressiveness [15,16]. The potential aggressiveness and recurrence are the main reason of the very challenging management of this tumor [20-45]. For now, for spinal CCMs, total resection should be considered as the optimal treatment. In our review of literature, 79 (89.77%) cases received Gross Total Resection (GTR) as the optimal treatment. However, 20 cases of spinal CCMs (22.72%) still experienced one or more recurrences until the last follow up. The use of postoperative radiotherapy has been increasingly affirmed and highlighted in the treatment of intracranial CCMs [3,21]. However, because of its rarity, the efficacy of postoperative radiotherapy for spinal CCM cannot be irrefutably concluded, therefore it requires further investigation. Tao et al. [21] reported that radiotherapy should not be performed immediately after the first operation of spinal CCMs, because the recurrence rate is lower than the one of intracranial CCM. This may be explained by the higher rate of GTR in spinal CCMs. According to the reported data, it is evident that spinal CCMs are more aggressive, with a much greater progression rate (38.0%) after initial treatment compared with ordinary spinal meningiomas (~3%) [13]. Jiu Hong Li et al. showed in their study that younger patients had better evolution after treatment (54.2%) compared with older ones (23.1%) [22, 46-53].

Conclusion

Spinal CCM, a rare subtype of meningioma, is an aggressive neoplasm with a high rate of local recurrence. It has a predilection for younger patients, and it is mostly located in the lumbar spine. Commonly, the spinal CCM is attached to the dura mater and it is exceptional to see a non-dura linked CCM. Gross total resection remains recommended in treating spinal CCM whenever it is possible. However, radiotherapy could be considered for patients who have undergone subtotal resection or for younger patients, regardless of the extent of resection [54-56].

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