Intramedullary Spinal Cord Cavernoma; A Case Report and Literature Review

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Abstract

Background: Spinal cord intramedullary cavernous malformation (SICM) is kind of rare vascular disease, and the therapeutic strategy is still under debate. Cavernous malformation (CM) can be found throughout the central nervous system (CNS) but only rarely occur within the spinal cord. The purpose of this article is to describe natural history, clinical presentation and outcome of SICM case which treated surgically

Case Presentation: A 70-year-old healthy woman presented with lower extremity weakness. A posterior laminectomy was performed, and a diagnosis of intramedullary spinal cord cavernous malformation was established. Neurological improvement was seen in one month after surgery

Conclusion: Intramedullary CM is a rare disease but one with significant consequences if not managed appropriately. whenever safely feasible, gross total resection is suggested, to prevent rebleeding and further worsened of neurological deficit.

Introduction

Cavernous malformations are well circumscribed vascular lesions made up of thin-walled sinusoidal channels.[1] Cavernous malformations (CMs) are not uncommon, but most of them are found to be located intracranially. Intramedullary CMs are rare, which comprise 3–5% of identified total central nervous system lesions and account for 5–12% of all spinal cord vascular lesion, with annual risk haemorrhage 2.1-2.5%.[2,3,4,5] Appropriate management of these lesions requires understanding their clinical, radiographic characteristics and a thorough knowledge of the anatomy of the spine and spinal cord.

Case Presentation

A 70-year-old healthy woman presented after an episode of back pain since the last 1 month, followed by progressive lower extremity weakness (motor strength 4/5) and dysesthesias. Magnetic resonance imaging demonstrated a T1W mix density, predominantly hyperintense lesion from T-2 through T-4 Differential diagnosed with an intramedullary spinal cord tumour haemorrhage or spinal intramedullary CM.

A posterior laminectomy was performed. After opening the dura we found bluish discoloration, then a posterior median sulcus myelotomy was performed over this area. We then exposed the hematoma, blood products were suctioned then we resected the lesion piece by piece along the gliotic plane, leaving the hemosiderin margin as a guide for the

cleavage plane of the lesion. Intraoperative somatosensory evoked potentials (SSEPs) and motor evoked potentials (MEPs) were monitored. After complete resection of the lesion, we sutured the pia mater, arachnoid membrane, and duramater.

No transient deficit postoperatively. One month after surgery, her motor examination was entirely normal but her dysesthesias persist.

Figure 1. Contrast Sagittal T1-weighted MR image demonstrating heterogeneous mixed signal intensity at T2-T4 with no enhancement

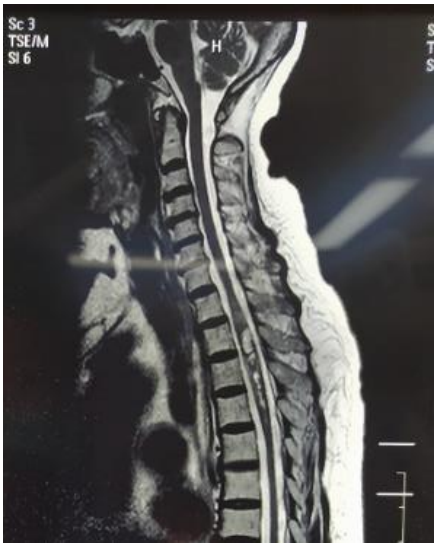

Figure 2. Sagittal T2-weighted MR image demonstrated mixed signal with near complete T2 hypointense rim at T2-T4

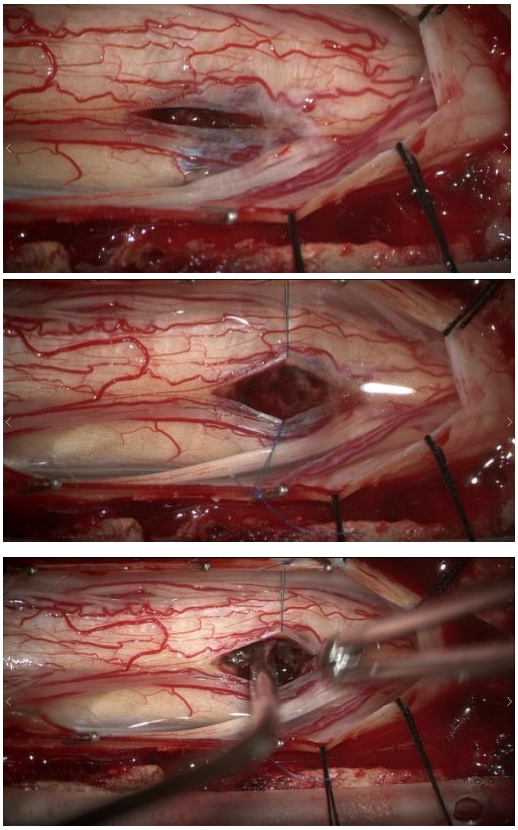

Figure 3. Posterior intramedullary cavernoma can be seen under the duramater. A small myletomy was made over the hemosiderin staining and the hematoma cavity was entered.

Discussion

Cavernoma is a vascular malformation, raspberry-like lesion filled with blood that flows very slowly through vessels that like “caverns”. The caverns is composed of dilated thin wall capillaries that consist of single layer of endothelial cells, very few intact tight junction between the endothelial cells and no elastic fiber. Cavernoma are dynamic lesions, some will remain stable without bleed for a very long period, some might bleed once than remain stable, some might grow, but some could bleed repeatedly. Patient with repeated bleeding will have progressive declining of neurological state. Although it is recovering, but the functional state never get back to the function prior to the last bleed. Complete removal of cavernoma is essential to prevent rebleeding.[1]

In line with our literature study, thoracic region is the most common site. On fourty one publications with 1032 patients of spinal cord cavernoma in the past 10 years.[2,3,6-47] (Table 1). The mean age at symptom onset was 41 years. The male/female ratio was nearly the same 1.24:1. Fifty nine percent (611 cases) of lesions were located at thoracic spine, 35% (368 cases) at cervical, 1% (11 cases) at lumbar region, 1.7 % thoracolumbar, 1.6 % cervicothoracal, 0.2 % conus medullaris 0.09% cervicomedullary, and 0.4 % unspecified location.

Although it was not found in our case, on our literature studies, we found 29 (9%) of 298 patients evaluated for a family history of these lesions had at least one family member with at least one CM.[3,6,7,8] Fifty four (38%) of 141 patients described in 4 series had synchronous intracranial CMs.[3,7,8,9].

Extremity weakness is the most common presentation, as seen in our case. It is consistent with 38 publications with 898 patients provided information on patient clinical presentations, 577 patients (64%) presented with motor weakness, 510 patients (56.8%) had sensory deficit, at least 358 patients (39.8%) had pain, and 320 patients (35.6%) had bowel or bladder dysfunction.

Table 1. Clinical data from series of SICM [6-47]

Authors & year	Patients	Mean Age (yr)	M: F Ratio	Lesion location No of lesion	Clinical presentation No of patients	Onset, No of patients	Family history
Bozinov, et al. 2011	6	27.5	3:3	Cervical 2, Thoracal 4	Weakness 4, sensory deficit 4, B/B 1	Acute 2, progressive 4	-
Choi,et al. 2011	21	39.3	8: 13	Cervical 9, Thoracal 9, Lumbal 1 Cervicomedullary 1 Thoracolumbar 1	Weakness 12, sensory deficit 11. B/B 4, pain 9	Acute 8, progressive 8, recurrent 5	-
Liang et al. 2011	96	34.5	1,67:1	Cervical 25 Thoracal 68 Lumbal 3	Weakness 84, sensory deficit 46 B/B 68, pain 39	Acute 6 Progressive 90	0
Pearl et al. 2012	2	52	2:0	Thoracal 2	Sensory deficit 2	Progressive 2	-
Savasta et al.2012	1	9	0:1	Thoracal 1	Pain 1	Acute 1	-
Soriasno et al. 2012	1	42	0:1	Thoracal 1	Sensory deficit 1 Pain 1	Acute 1	-
Tong et al. 2012	20	34	1:3	Cervical 9 Thoracal 7, Cervicothoracal 4	Weakness 14, Sensory deficit 9, pain 5, B/B 2	Progressive 20	-
Babu et al. 2013	8	43	-	Thoracal 4 Others 4	Weakness 6 , sensory deficit 6, pain 5, B/B 1	Progressive 8	-
Eicker et al. 2013	1	20	0:1	Cervical 1	Sensory deficit 1 Pain 1	Progressive 1	-
Endo T et al. 2013	8	57	5:3	Thoracal 8	Weakness 2, sensory deficit 2, pain 4	Acute 5 Progressive 3	-
Lanotte, et al 2013	1	24	0:1	Cervical 1	Sensory deficit 1	Progressive 1	-
Grasso et al. 2014	1	34	0:1	Thoracal 2	Weakness 1 sensory deficit 1 , B/B 1	Acute 1	-

Matsui et al.2014	1	42	1:0	Conus medullaris 1 Skin associated lesion 1	Weakness 1 sensory deficit 1 , B/B 1	Acute 1	-
Qing et al. 2014	20	48.5	8: 12	Cervical 9 Thoracal 11	Pain 20	Progressive 20	-
Ardeshiri et al, 2015	25	46	14: 11	Cervicothoracal 11 Thoracolumbar 14	weakness 16, sensory deficit 21 B/B 4	Acute 1 Progressive 16 Recurrent 3	-
Reitz M et al, 2015	48	41.3	25:23	cervical 19 thoracic 27 thoracolumbar 2	Asypmtomatic 3	Acute 20 Progressive 9 Recurrent 16	-
Zhang et al. 2016	85	40.5	51:34	cervical 40 thoracal 40 lumbar 5	weakness 62 sensory deficit 67 pain 33 B/B 20	Acute 41 Progressive 40 Recurrent 4	-
Kodeeswaran et al, 2016	1	15	1:0	Cervical 1 Synchronous intracranial lesion 1	Weakness 1	Progressive 1	-
Scherman et al, 2016	1	56	0:1	Cervical 1	Weakness 1 Sensory deficit 1 B/B 1	Progressive 1	-
Salinas et al, 2017	1	62	1:0	Thoracal 1	Weakness 1 Sensory deficit 1	Progressive 1	-
Ahmed NF, et al, 2017	1	45	1:0	Cervical 1	Weakness 1, sensory deficit 1, Pain 1	Progressive 1	-
Huntley et al. 2017	1	58	1:0	Thoracal 1	Weakness 1, Sensory deficit 1, B/B 1	Progressive 1	-
Imagama S, et al, 2017	41	39	18:23	Cervical 17 Thoracal 24	Weakness 26	-	-
Azad TD et al. 2017	32	44.2	13:19	Cervical 16 Thoracal 16 Synchronous intracranial lesion 9	Weakness 5 sensory deficits 9 Pain 16 B/B 2	Acute 19 Progressive 13	6
Sun I et al.2017	10	45	1:1	Cervical 6 Thoracal 4	-	Acute 4 Progressive 6	-
Li et al. 2018	83	39	40:43	Cervical 34 Thoracic 47 Lumbar 2	Weakness 47 Sensory deficits 9 Pain 25 B/B 13	Progressive 83	6
Nagoshi et al. 2018	2	37.5	2:0	Cervical 2	Weakness 1 Sensory deficit 1 Pain 1	Acute 2	-
Oh HM et al.2018	1	79	0:1	Cervicothoracal 1	Weakness 1 Pain 1 Sensory deficit 1	Acute 1	-
Ren et al.2018	10	39.9	5:5	Cervical 2 Thoracal 8	Weakness 5 Sensory deficit 7 Pain 7 B/B 3	Progressive 10	-
Winkler AE et at. 2018	1	28	1: 0	Thoracal 1	Weakness 1 Conus medullaris syndrome 1	Acute 1	-
Apostolakis et al. 2019	1	77	1:0	Conus medullaris 1	Pain 1	Progressive 1	-
Goyal et al. 2019	107	17.3	59:48	Cervical 43 Thoracal 63 Conus medullaris 1 Synchronous intracranial lesion 24	Motor 14 Sensory 16 Pain 17 B/B 15 Asymptomatic 22	Acute 28 Progressive 56	11

Kang MG, et al. 2019	1	61	1:0	Thoracal 1	Weakness 1 Pain 1	Progressive 1	-
Moldovan K, et al. 2019	1	54	1:0	Thoracal 1	Motor weakness 1	Acute 1	-
Ren J et al. 2019	20	14	16: 4	Cervical 5 Thoracal 15	Weakness 19 Sensory deficit 20 Pain 10 B/B 12	-	-
	234	38.3	132: 102	Cervical 62 Thoracal 172	Weakness 197 Sensory deficit 224 Pain 133 B/B 145	-	-
Ren J et al. 2019	40	40.6	2.1:1	Cervical 17 Thoracal 23	weakness 34 sensory deficits 37 pain 16 B/B 17	Acute 36 Progressive 4	-
Couldwell WT et al. 2020	1	36	1:0	Cervical 1	Weakness 1	Progressive 1	-
Gendle C et al 2020	1	5 mo	1:0	Thoracolumbar 1	B/B 1	Progressive 1	-
Nwachuku et al. 2020	1	63	1:0	Cervical 1	Weakness 1 Sensory deficit 1 B/B 1	Progressive 1	-
Oishi M et al 2020	1	37	1:0	Cervicothoracal 1	Weakness 1 Pain 1 B/B 1	Acute 1	-
Panda A et al. 2020	76	49	39:37	Cervical 35 Thoracal 41 Synchronous intracranial lesion 20	Asymptomatic 17	Acute 9 Progressive 29 Recurrent 21	6
Zhang et al. 2020	18	12.9	2: 1	Cervical 9 Thoracal 9	Weakness 15 Pain 10 Sensory deficit 8 B/B 6 Spinal deformity 1	Acute 11 Progressive 7	

Although the clinical presentation may be variable, the majority of patients seek medical attention due to acute onset of hemorrhage into the spinal cord parenchyma as seen in aour patient. Ogilivy et al describe four types of clinical presentation. First, acute neurological deterioration over a period of months or years with gradual, partial improvement in between episodes. This probably the result of episodes of small hemorrhages from the lesion. Second, slowly progressive neurological deterioration which lasted several months to years. This suggests an enlargement of the lesion resulting from repeat microhemorrhages, the gliosis neurotoxic effect of hemosiderin, impaired microcirculation due to local pressure, or progressive hyalinization and thickening of the vascular walls or gradual thrombosis. Third, acute onset of neurological deterioration with a rapid neurological decline. Fourth, acute onset of pain and mild symptoms of neurological deterioration with gradual decline over weeks to months to the final syndromes.[48]

Magnetic resonance imaging is the gold standard for diagnostic. A typical lesion on an MR image is a lobulated, commonly said as popcorn appearance. Internal blood-fluid levels on either axial or sagittal sequences can also be found. Hemosiderin deposition resulting in a hypointense rim around a mixed signal intensity is the pathognomonic appearance of CMs on T2w. Adjacent intramedullary hemorrhage can also be founded separately from T2w hypointense rims.[8,12]

Based on the signal characteristics, SCMs were also classified into 4 types according to Zabramski's criteria Type I (subacute hemorrhage) if it was hyperintense on T1w and hypo/hyperintense on T2w with an incomplete or absent T2w hypointense rim. Type II ("classic") if it was heterogeneous on both T1w and T2w with complete/near complete Tw2 hypointense rim. Type III (chronic hemorrhage) if it was isointense/hypointense on T1w and T2w with varying extent of T2w hypointense rim. It was considered type IV (punctate hemorrhage) if it was not well seen on T1w and T2w.[8,49]

Cavernous malformations must remain in the differential diagnosis of intradural intramedullary lesions. The broader differential includes multiple sclerosis, spinal ependymomas, astrocytomas, metastatic disease, hemangioblastomas, spinal AVMs, and transverse myelitis. Spinal angiography will facilitate the diagnosis of a spinal vascular malformation.[12] Moreover, in particular case, spinal angiography enhanced with flat-panel catheter angiotomography (FPCA) is suggested to identify spinal developmental venous anomaly in cavernous malformations to prevent higher risk of surgical complication.[13]

Resection is the only definitive treatment since this leads to a permanent elimination of the risk for further growth or hemorrhage. The resection of the active cavernoma tissue is thought to be sufficient. Excision of hemosiderin infiltration in the surrounded healthy tissue is not recommended, since it poses a significant risk of morbidity. Incomplete resection may lead to recurrence and even progressive deterioration due to bleeding from the residual malformation.[10,12]

Across 23 literature studies with 825 patients, 671 CMs (81%) were totally resected.[3,7,9,10-12,16,17-19,24-26,28,29,32,34,36-38,41-43] Transient early postoperative morbidity was reported in 15 out of 116 patients (12%) in 3 series, At the long-term follow-up 224 patients were improved (27.15%), 393(47.5%) were unchanged, and 72 (8.7%) were worsened, as compared with their preoperative status. These overall results are summarized in Table 2.

Table 2. Outcome on surgically treated SICM [3,7,9,10-12,16,17-19,24-26,28,29,32,34,36-38,41-43]

Authors & year	No of patients	Total resection rate	Early transient morbidity	Mean FU in months	Long term outcome No of patients
Bozinov, et al. 2011	6	6/6			Improved 6
Choi,et al. 2011	21	21/21		33.21 (1-73)	Improved 10 same 9 worsened 2
Liang et al. 2011	96	64/96	5	45.8 (10–183)	Improved 23 Same 35 worsened 6
Tong et al. 2012	20	20/20	8		Improved 7, same 5
Babu et al. 2013	8	8/8		15.9 (36.7 ± 10.4)	Improved 4, same 2, worsened 2
Eicker et al. 2013	1	1/1		3	Improved 1
Endo T et al. 2013	8	8/8		28.9 (15 to 52)	Improved 5 same 2 worsened 1
Ardeshiri et al, 2015	25	20/25		6.3 (2-17)	Improved 2, same 18
Reitz M, 2015	48	48/48		79.3 ± 35.2	Improved 6 Same 26 Worsened 16
Zhang et al, 2016	85	58/85		42.8	Improved + same 79 : Worsened 6
Kodeeswaran et al, 2016	1	1/1		6-8	Improved 1
Scherman et al, 2016	1	1/1		11	Improved 1
Salinas et al, 2017	1	1/1		10	Improved 1
Imagama S, et al, 2017	41	41/41		10 years (2-24 years)	Improved 11 Same / worsened 15
Azad TD et al. 2017	32	32/32	2	6	Improved 6 Same + worse 20
Li et al. 2018	83	82/83		0.5-6 years	Improved 19 same 38 worsened 5
Oh HM et al. 2018	1	0/1		3	Improved 1

Ren et al.2018	10	10/10	1.4 years (3 mo – 6.8 mo)	Improved 2 same 8
Winkler AE. Et at. 2018	1	1/1	7 week	Improved 1
Goyal et al. 2019	107	32/107		Improved 10 Same 15 Worsened 7
Moldovan K, et al. 2019	1	1/1	6 week	Improved 1
Ren J et al. 2019	15	15/15	42.7 ± 36.8 (6 - 164 mo)	Improve 5 Same 9 worsened 1
	176	176/176	46.4 ± 34.6 (6 - 164 mo)	Improve 57 Same 113 Worsened 6
Ren J et al. 2019	37	24/ 37	40.5 ± 38.9 months	Same 17 Improved 5 Worsened 2

Gross-total resection is the most important factor in eliminating the risk of a new haemorrhage in the future, It is achieved in 94% The transient worsening of patients immediately following the surgery is seen in 24–50% cases and the majority resolves during the first 3 months.[4,5] Another study reported on 7 % of patients which suffered from neurological worsening that unimproved during the long-term follow-up. Only a minority of published studies have reported results of conservatively treated patients.[12] Based on these studies surgical resection is the chosen way of treatment for our patient.

Intramedullary cavernomas may be located superficially or deep-seated; anteriorly, laterally, or posteriorly. Posterior approach is the most frequently elected, because it's provide extensive exposure of the spinal cord, relatively safe, and furthermore, it is an approach which all neurosurgeons are familiar with. A lateral lesion can be approached posteriorly by releasing dentate ligament to allow cord rotation to perform direct entry to the lesion. For a centromedullary lesion without any surface discoloration, a myelotomy should be performed during the operation, which might involve a higher surgical risk. Furthermore, SSEP and MEP monitoring is useful in protecting neurological function.

Length of symptoms and pre operative neurological state is an important factor predicting postsurgical recovery. In long-term follow-up, Svoboda et al reported 42% improved (11 patients), 46% remained stable (12 patients), and 12% (3 patients) deteriorated compared to their presurgical neurological status.[50] It is general agreement that patients with motor deficits have the highest possibility of postsurgical improvement reaching up to 86%.[50] Interestingly, several studies pointed out that sensory deficit, pain (radicular or central), and bladder dysfunction often do not favor a full recovery following the surgery.[4,5,31] Patients having symptoms of less than 3 months were found to have better postsurgical outcomes Similarly, patients with acute or stepwise clinical course are prone to benefit more significantly from surgery when compared to those with progressive neurological decline.

Conclusion

Spinal cord intramedullary cavernoma are rare vascular malformation of the spinal cord. Removal of intramedullary spinal cavernomas may lead to clinical improvement, so it may be considered as a curative option to reduce the risk of new hemorrhage. We would consider surgical removal of symptomatic intramedullary spinal cavernomas with proven growth or bleed with significant initial symptom.

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