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Case Report

Intradural Extramedullary Capillary Hemangioma of Spinal Cord: A Case Report and an Updated Review

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ABSTRACT

Background and Aim: Capillary hemangiomas are benign tumors found on the skin and soft tissues. They rarely present as an intradural spinal tumor. Common differential diagnosis methodsareschwannoma, hemangioblastoma, metastasis, and paragangliomas.

Case Presentation: We report a case of a 38-year-old female with complaints of lower backache with radiation to lower limbs, in which the magnetic resonance imaging revealed an intradural tumor compressing the cauda equina nerve roots, arising from the L3 level. The patient underwent L2-L3 laminectomy with tumor excisionwith the preservation of nerve roots.

Conclusion: Histopathology suggested capillary hemangioma and the patient improved symptomatically and no recurrence has been reported todate.

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Highlights

- Spinal intradural extramedullary capillary hemangiomas are rare.
- Capillary hemangiomas are benign lesions causing symptoms by compression of nerve roots.

Plain Language Summary

Capillary hemangiomas are benign tumors that appear on the skin and soft tissues. They are rarely found in the spinal area. Doctors need to consider several possible causes when facing a tumor in the spine area. This includes schwannoma, hemangioblastoma, metastasis, and paraganglioma. We examined a 38-year-old woman complaining about lower back pain radiating down to her right thigh. We did amagnetic resonance imaging of her lower spine and found a tumor near the L3 level that was putting pressure on the nerve roots. We thought it was a benign nerve sheath tumor; therefore, we performed surgery to remove the tumor without harming the nerve roots. We found that the tumor was a capillary hemangioma based on a test of its tissue.The patient's symptoms improved after the surgery and we followed up her for 6 months to ensure that the tumor did not return.

1. Background and Importance

Primary spinal cord tumors are less common compared with primary tumors of the brain. They constitute only 2% to 4% of all primary central nervous system (CNS) tumors [1, 2]. Common spinal intradural tumors are benign nerve sheath tumors, meningioma, and ependymoma. Capillary hemangiomas in the spinal location are rare, with only about 21 cases reported worldwide. Capillary hemangiomas are believed to be hamartomatous proliferations of vascular endothelial cells. The surgical implication of this is the risk of excessive bleeding during surgery, which can lead to adverse outcomes. Only a few cases have been reported as intradural capillary hemangioma [3, 4]. We are reporting a case of a 38-year-old womanwith capillary hemangioma in the cauda equine region. This study aims to present a detailed and documented analysis of this particular case, including symptoms, diagnosis, treatment, and outcome. Capillary hemangiomas should be kept as rare but plausible differentials in the spinal intradural extramedullarywith the potential for intra-operative excessive bleeding.

2. Case Presentation

A 38-year-old woman came with majorcomplaints of low backache for 4 months. The pain was lancinating in nature. It was radiating to the right thigh and leg along with numbness and weakness in the right foot. The pain was increasing during the night and was not relieved by medicine. The intensity of the pain gradually increased with time. In physical findings, the straight leg raising test was positive on the right side. She also presented with left ankle weakness and sensory deficits in the left L3-L4 dermatome. No local spinal tenderness was observed.

Contrast-enhanced magnetic resonance imaging (MRI)of the lumbosacral spine showed a well-defined lobulated lesion in the spinal canal regional L2-L3 level, in thecal sac, in an intradural compartment with dimensions as 18x14x11 mm, which was isointense on T1 weighted image and slight hyperintensity on T2 weighted image (Figure 1). There was a displacement of nerve roots with gadolinium contrast homogenous enhancement. A provisional diagnosis of benign nerve sheath tumor was put forward. The whole-brain MRI screening was done to rule out intracranial lesions. The rest of the routine investigations were within normal limits except for blood sugar and HbA1c-7.9%. The patient was newly diagnosed with diabetes mellitus type 2 and started on appropriate oral hypoglycemic drugs by the endocrinology team.

The patient underwent L2-L3 laminectomy with tumor excision under general anesthesia in a prone position. A reddish-brown tumor was visualized intraoperatively on midline durotomy at the corresponding level. The tumor was seen compressing underlying nerve roots. With fine dissection, the tumor was resected out and was found to be originating from a single nerve root which had to be sacrificed for complete tumor excision. After achieving complete hemostasis, primary dural closure was done with prolene 4-0.

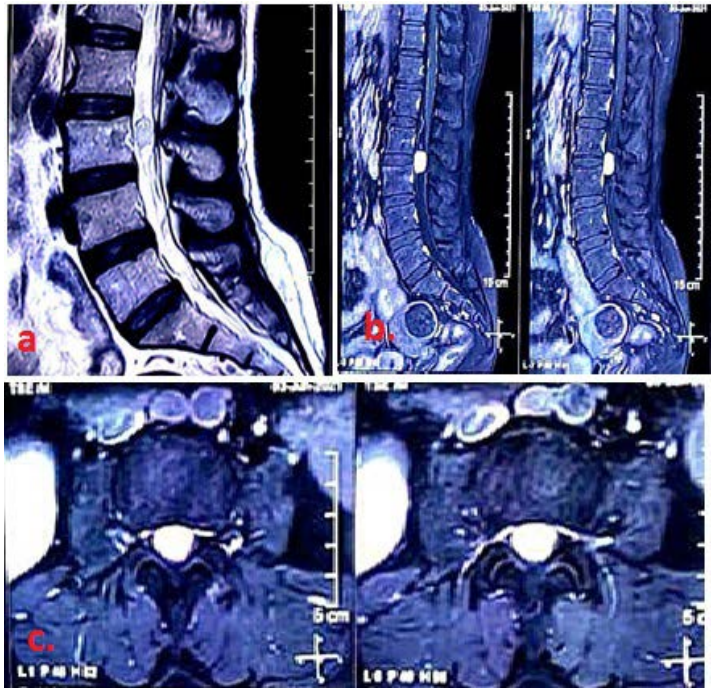

Figure 1. Intradural mass lesion

a) Hyperintense onT2 image;

b) Homogenous enhancement on a post-contrast image, measuring approximately 12x9x14 mm seen at L3 level;

c) On axial images displacing caudate equine nerve root toward left and posterior.

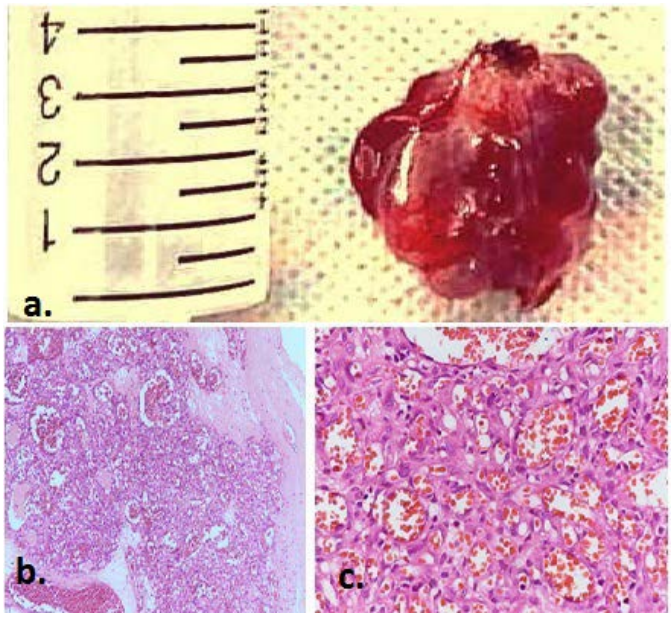

Figure 2. Histopathology of Lesion

a) Gross appearance of the tumor;

b) Histopathology slide showing circumscribed lesion composed of closely grouped proliferating capillary channels (H&E x100);

c) A lack of atypia or mitotic activity (H&E x400).

Table 1. Previous case reports on intradural extramedullary capillary hemangiomas

Author	Age (y)/Sex	Site and Size	Clinical Features	Treatment	Prognosis
Chung et al. 2010 [13]	47/Male	T6-7 1x1.3x1.5 cm	bilateral radiculopathy, sensory impairmentbe-low T7, increased deep tendon reflexand ankle clonus	Complete removal of the mass	Gradual sensory improvement
Miri et al. 2009 [14]	20/Male	L3	Low back pain and cauda equine syndrome	En bloc excision of mass	Weakness im-proved, urogenital symptoms margin-ally improved
Shin et al. 2000 [15]	66/Female	T8-9 1.3x2 cm	Grade 3 paraparesis	Incomplete excision due to significant bleeding	Marked neurologi-cal improvement, magnetic reso-nance imagingat 6 months–no residual tumor
Abdullah et al. 2004 [16]	32/Male	T10 1.7x1.4x1 cm	Low back pain, weakness of the left>right limb weakness, sensory loss below T9	In toto excision	Gradual neurologi-cal improvement, no recurrence
Hanakita et al. 1991 [17]	58/Male	L1-2	Low back pain with cauda equina syndrome	Excision	Significant im-provement
Nowak et al. 2000 [18]	63/Female	T12-L1	Low back pain with hypoesthesia	Mass excision	Residual paresis, no recurrence
Yu et al. 2006 [19]	48/Male	T6-7 1x1x0.7 cm	Sudden onset of back pain with paraparesis and hypoesthesia	Total removal along with dorsal root	Neurological improvement, no recurrence
Ghazi et al. 2006 [20]	42/Male	L3-4 2x1.5 cm	Low back pain with right lower limb radiculopathy	Tumor and nerve root removal	Symptoms resolved completely, mild plantar flexion weakness
Andaluz et al. 2002 [21]	41/Male	Conusmedullaris 2x1 cm	Low back pain with radiculopathy, absent knee reflex	Excision	Neurologically recovered, no re-currence
Alkandy, 2006 [22]	60/Male	T9	Low back pain, parapa-resis, numbness in the right limb and below the left knee	Total excision	Neurologically improved
Funayama et al. [23]	34/Male	L4 1x1x1 cm	Low back pain, absent, sensory loss over the left foot	Mass was excised	No recurrence
Choi et al. 2001 [24] Case1 Case2 Case3	28/Male 53/Male 51/Male	L1, 1.5x1x1 cm T5-6 1.5x0.5x0.5 cm 2x1x1 cm	Low back pain with paraparesis Claudication, paraparesis, and hypoesthesia Claudication and radiat-ing pain to both limbs	Mass was excised	Not commented
Bozkus et al. 2003 [25] Case1 Case2	55/Male 37/Female	T8 (intra+extramedullary) T5-6(intra+extramedullary)	Bilateral leg numbness, decreased sensation below T12 Bilateral leg numbness, flaccid paraparesis, urinary retention	Two-stage surgery for extra-medullary and intramedullary part Complete exci-sion	Complete excision small residual intradural at 2-monthmagnetic resonance imaging

Histopathological examination of the specimen revealed a well-circumscribed tumor composed of capillaries arranged in the lobular configuration without atypia or mitotic activity suggestive of capillary hemangioma (Figure 2). The patient was followed up in an outpatient setup with serial MRI studies and clinical examinationand showed improvement in symptoms and no recurrences todate.

3. Discussion

Histological features of lobular capillary hemangioma are the presence of normal-sized (size of the capillary) channels tightly aggregated into nodules, each supplied by a feeding vessel [5].

Other than the usual locations of capillary hemangiomas, i.e. cutaneous and subcutaneous, they are rarely seen in neuroaxis [6, 7]. Frequently reported capillary hemangiomas are dural [8], peripheral nerves, muscles [5], skin [9], and mucous membrane [10]. So far, only 21 cases reported as per an advancedsearch on PubMed regarding intradural spinal capillary hemangiomas of the cauda equine region [11]. Hemangiomas may arise from blood vessels of a nerve root in the cauda equine, an inner surface of the dura, or the pial surface of the spinal cord, and the adherence of other roots reflects its associated arachnoiditis [12]. Capillary hemangioma presents as a space-occupying mass leading to chronic progressive myelopathy or radiculopathy and sensorymotor deficits with pain. Sudden neurological deterioration can occur due to the risk of bleeding. We achieved a gross total resection of the tumor in our patient with no recurrences todate. This is at par with the current literature evidence briefly discussed in Table 1.

4. Conclusion

Capillary hemangiomas are rare in intradural extramedullary space with clinical and radiological similarities with common spinal lesions.Therefore,they should be considered a differential diagnosis before surgical intervention.

Ethical Considerations

Compliance with ethical guidelines

Written informed consent was obtained from the patient.

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Authors' contributions

All authors contribute to the design, running, and writing of all parts of the research.

Conflict of interest

The authors declared no conflict of interest.

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