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CASE REPORT

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Extradural lumbosacral cavernous hemangioma

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Abstract Purely extradural cavernous hemangiomas of the spinal canal are extremely rare. Their occurrence at the lumbosacral level is a true exception. We describe a case of lumbosacral extradural hemangioma and review the behaviour and MR imaging characteristics of these lesions. Cavernous hemangioma should be considered in the differential diagnosis of an enhancing extradural periradicular mass causing chronic lumbar radiculopathy especially in fertile women. Lumbar extradural hemangiomas appear and behave differently, not only from intramedullary cavernous hemangiomas but also from extradural hemangiomas of other spinal locations.

Keywords Cavernous angioma · Epidural space · Lumbar disk disease · MRI

Introduction

Pure extradural cavernous hemangiomas of the spinal canal are extremely rare. This lesion in this specific location was first described by Globus and Doshay in 1929 [4]. Seventy-six cases have been reported for the whole spinal canal and in the majority of these cases the tumor had a predilection for the upper thoracic spine [2, 14]. Only 11 cases of extradural cavernous hemangioma involved at the lumbar or lumbosacral spine exist according to the literature [1, 3, 6, 7, 8, 9, 11, 13, 15, 18]. The symptoms and signs caused by lumbar or lumbosacral extradural cavernous malformations are often quite insidious and vague and radiculopathy caused by such a lesion is often attributed to disk herniation. The aim of this article is to present a rare case of low back pain and sciatica caused by extradural cavernous hemangioma, to briefly review the literature, and to highlight the diagnostic challenges associated with these lesions.

Case history

A 28-year-old woman presented with 2-year history of back pain and ten-month history of right leg pain. The leg pain got worse with coughing and straining. During her pregnancy that took place 4 years prior to presentation, she experienced severe low back pain especially during the last trimester. The pain eased to tolerable levels immediately after childbirth. On examination, the power and the deep tendon reflexes in the legs were normal, but sensation to

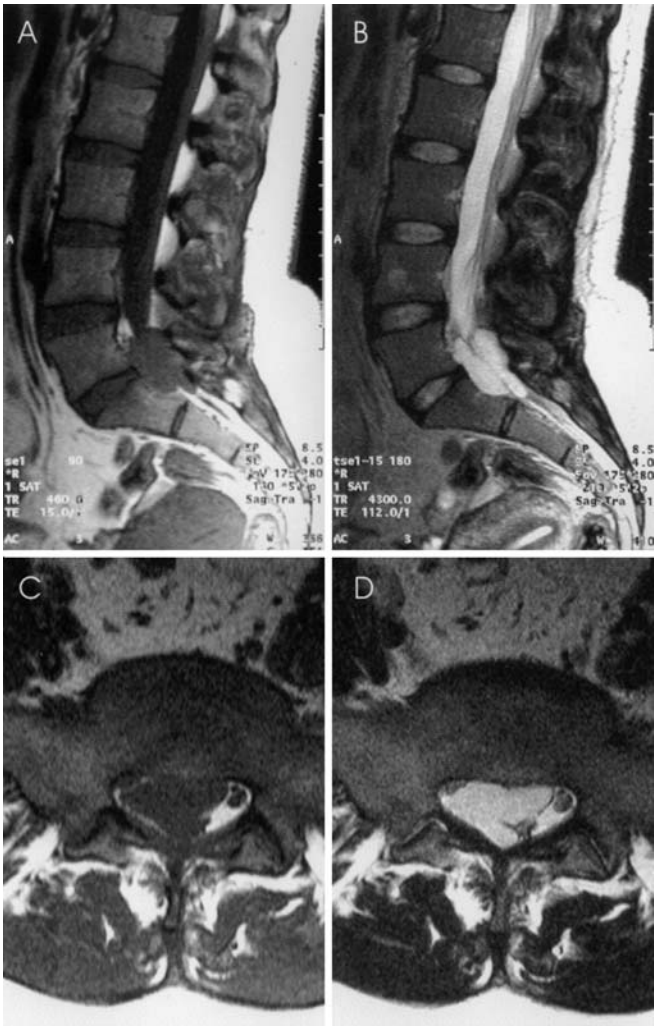

Fig. 1a–d The MRI demonstrates a 4×3×2-cm lobulated lesion that envelopes the distal dural sac. The lesion is **a, c** isointense on T1-weighted image, **b** hyperintense on sagittal T2-weighted image, and **d** homogenously enhanced on axial T1-weighted image

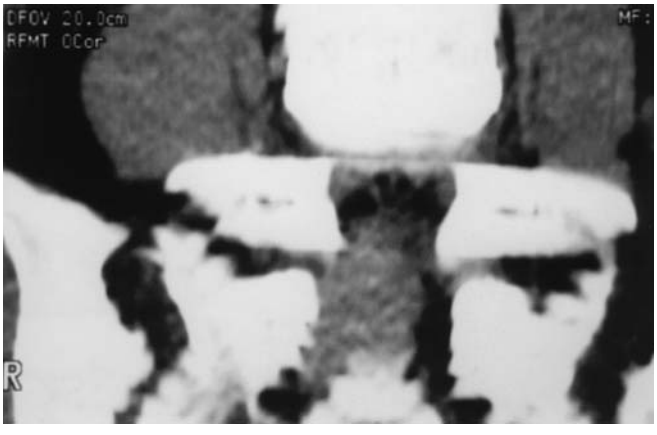

Fig. 2 Oblique coronal contrast-enhanced CT. There is a downward extension of predominantly right-sided lumbosacral mass into a normal-sized right S2 neural foramen

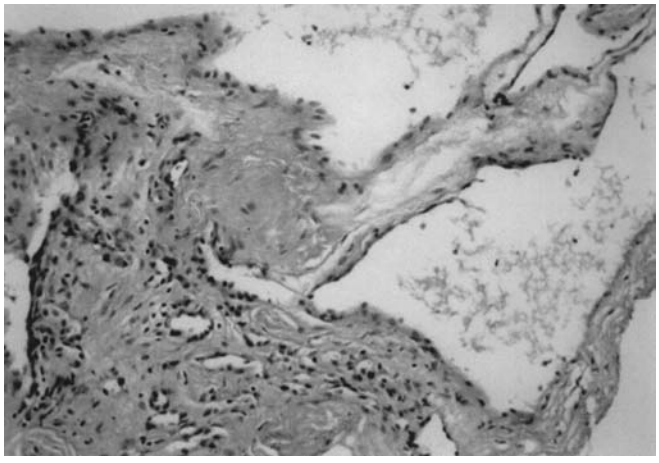

Fig. 3 Photomicrograph of the extradural mass shows endothelium-lined cavernous channels supported by a stroma, relatively rich in cellular content, characterizing a cavernous hemangioma (hematoxylin and eosin, ×200)

pinprick was decreased over the right S1 dermatome. The straight leg-raising test was positive at 60° on the right side with significant bilateral paravertebral muscle spasm. Magnetic resonance imaging (MRI) with a 1.5-T unit revealed a lobulated extradural mass at L5–S1 level that filled the whole canal and measured 4×3×2 cm. The mass was isointense (with the spinal cord) on T1-weighted MR images and hyperintense on T2-weighted MR images (Fig. 1a,b). The lesion was encircling the distal dural sac and the right S1 nerve root (Fig. 1c). Part of the lesion was extending inferiorly through the right S2 foramen. The mass showed marked homogenous enhancement (Fig. 1d). Fat-suppressed spin-echo T1-weighted as well as fat-suppressed T2-weighted MR images did not show any lipomatous signal within the lesion. Computed tomography (CT) of the lumbosacral spine was done next to better delineate the distal extension of the extradural lesion and to measure and compare the width of sacral nerve root foramina. The CT showed normal size of sacral nerve root foramens and better delineated the infero-lateral extension of the mass (Fig. 2). The preoperative diagnosis of extradural hemangioma was made and angiogram was deemed unnecessary.

The lesion was approached through L5 and engorged upper sacral canal laminectomy. The lesion appeared as a dark red encapsulated mass. It was with its vascular supply stemming from the vessels originating from the posterior longitudinal ligament. The lesion was moderately adherent to dural sac and the right S1 nerve root sleeve. Using the microsurgical technique, it was possible to strip the lesion from the dural sac, and to excise it en bloc without damaging the dura.

Histopathological diagnosis was cavernous hemangioma (Fig. 3). Sections revealed closely opposed vascular channels lined with endothelium without any neural tissue in between. Histology did not reveal any sign of an old or a recent hemorrhage or a microhemorrhage. Imaging of the whole neuroaxis failed to reveal another hemangiomatous lesion [17]. At 3 years postoperatively, the patient was well with occasional mild back pain. Follow-up MR scans have not shown recurrences.

Discussion

Vertebral body hemangiomas are known for their capability to extend into the extradural space[16], but by definition purely extradural cavernous hemangiomas of the spi-

Table 1 Summary of reported lumbar extradural cavernous hemangioma cases. *GRE* gradient-echo image, *LBP* lower back pain, *LP* leg pain, *NA* not available, *NG* contrast material was not given, *PD* proton-density image

Reference	Age (years)/ gender	Symptoms	Level	Location/shape	MR intensity		Contrast enhancement	Surgery/outcome
					T1-weighted MR	T2-weighted MR		
[3]	50/M	LBP+right LP	L3–L4	Right lateral/round	Hypo/isointense	Hyperintense	Peripheral	Total excision/no follow-up
[8]	70/M	LBP+left LP	L4	Left lateral	MR not done			Total excision/stable condition at 2 years
[7]	37/F	LBP+left LP	L3–L4	Left periradicular/ ovoid	Isointense	Hyperintense (GRE)	Heterogenous	Total excision/no follow-up
[11]	62/F	LBP+left LP	L2–L3	Left lateral/round	Isointense	Heterogenous (PD)	Not enhanced	Total excision/no symptoms at 1 year
	86/M	NA	L1–L2	NA	Isointense	Heterogenous	NA	Laminectomy+?/no symptoms at 1 year
[18]	44/F	Right LP	L4–L5	Right postero-lateral/ round	Isointense	Hyperintense	NG	Total excision/no symptoms at 18 months
[1]	26/F	Right LP	L5–S1	Right periradicular/ round	Hypointense	Hyperintense	NG	Total excision/persistent symptoms at 3 years necessitated embolization
[13]	51/F	LBP+right LP	L3–L4	Left periradicular/ round	Isointense	Hyperintense	Homogenous	Excision (total?)/mild symptoms at 3 years
	16/M	Left LP	L5–S1	Left ventrolateral/ ovoid	Isointense	Hyperintense	NG	Total excision/no symptoms at 5 years
	19/F	Left LP+pollakuria	L4	Left periradicular/ round	Isointense	Hyperintense	Not enhanced	Total excision/no symptoms at 1 year
[15]	19/F	Left LP+left hip pain	L4–L5	Left periradicular/ ovoid	Isointense	Hyperintense	Heterogenous	Total excision/mild symptoms at follow-up (follow-up with MRI)
[16]	28/F	LBP+right LP	L5–S2	Right periradicular and peridural/ globular	Isointense	Hyperintense	Homogenous	Total excision/minor symptoms at 3 years (follow-up with MRI)

nal canal are those that originate solely in the extradural space and that do not invade osseous structures. This subset of hemangiomas are well delineated and often have a capsule (or a pseudo-capsule). Their presentation is often due to compression (like a tumor) rather than vascular complications of intraaxial cavernous hemangiomas. Of all the occasional cases reported in the literature, a significant majority of purely extradural lesions occurred in the upper thoracic or thoracolumbar segments where presentation and diagnosis often takes place in an acute manner due to cord and/or conus compression [5, 18]. The extradural hemangiomas of the cervical [5] and lumbar and/or lumbosacral region appear to be the rarest of all.

Sudden onset of symptoms with serious neurological deficits of thoracic (or cervical) extradural hemangiomas have been related to enlargement or engorgement of the tumor. The etiology of neurologic deterioration is attributable to microhemorrhages within the matrix of the hemangioma, venous thrombosis, or venous outflow obstruction within the malformation. Interestingly, none of these catastrophic changes were reported for lumbar extradural hemangiomas which often caused back pain and ipsilateral leg pain [1, 3, 6, 7, 8, 9, 11, 18] and occasionally caused contralateral leg pain [13], ipsilateral hip pain [15], or pollakuria (Table 1) [13].

Magnetic resonance imaging revealed that hemangiomas appeared as intermediate (hypo- to isointense lesion) on T1-weighted and consistently hyperintense on T2-weighted MR images and with few exceptions these lesions exhibited homogenous contrast enhancement (Table 1). Previously reported extradural hemangiomas of the lumbosacral region consist of round or ovoid lesions located in the ventral and/or ventrolateral extradural space or just the periradicular space [8, 18]. Extraforaminal extension was exceedingly rare [7]. Our review of the literature revealed that the diagnosis of a lumbar extradural hemangioma was never made preoperatively. Even though a histological diagnosis from radiological images cannot be predicted with accuracy for each and every case, there are enough clues for differential diagnosis of an extradural lumbosacral solid lesion the imaging characteristics of which are summarized in Table 2. Briefly, differential diagnosis include schwannoma, neurofibroma, angiolipoma, lymphoma, metastasis, snovial cyst, osteochondroma, and disc herniation [6, 10, 13, 15, 18].

Complete removal of extradural cavernous hemangioma is the procedure of choice and usually bleeding is under control when operation is performed with microsurgical technique. The pseudo-capsule that these extradural lesions have often help the surgeon for a total excision. Significant bleeding may occur if the integrity of this capsule is breeched [13]. A total excision was possible in all but one of the previously reported purely extradural lumbosacral hemangioma cases. The literature data also shows that once totally removed, these tumors did not recur during follow-up periods of up to 5 years. Even when a complete removal is accomplished, symptoms may persist which may be due to scarring around the dural sac or the involved nerve root and do not necessarily point to a recurring lesion [15]. Endovascular treatment has been used in such cases to alleviate persisting symptoms with successful results [1]. Adjunct radiotherapy has also been advocated for incompletely removed extradural angiomas [9], but because radiotherapy is not without risk, we sug-

Table 2 Differential diagnosis of a lumbar extradural lesion based on MR and CT findings

	T1-weighted MR	T2-weighted MR	Enhancement with Gd-DTPA	CT with bone window
Benign group				
Cavernous hemangioma	Iso- or hypointense	Hyperintense	Homogenous enhancement	Normal-sized neural foramen (usually)
Disc herniation	Iso- or hypointense	Hyperintense (rarely isointense)	None or peripheral enhancement	Normal-sized neural foramen (usually)
Schwannoma	Iso- or hypointense	Hyperintense	Heterogenous enhancement with a rim	Enlarged neural foramen
Neurofibroma	Iso- or hypointense	Hyperintense	Heterogenous enhancement	Enlarged neural foramen
Angiolipoma	Hyperintense	Hyperintense	Homogenous enhancement (usually)	Normal-sized neural foramen
Osteochondroma	Hypointense	Hyperintense core with intermediate signal rim	None or minimal enhancement	Calcified mass within the foramen
Synovial cyst	Hypointense	Hyperintense	None or peripheral enhancement	Mass adjacent to facet joint; no bony change
Malignant group				
Metastasis				
Lymphoma	Iso- or hypointense	Inhomogenously hyperintense	Marked and homogenous enhancement	Bony destruction
Chordoma				
Ewing's sarcoma				

gest that such patients should instead be followed with frequent MRI assessments. A second-look surgery is clearly less risky than radiation.

Conclusion

Extradural cavernous hemangioma should be kept in mind as a remote differential diagnosis in a patient with chronic lumbar radiculopathy with MRI scans demonstrating an isointense lesion on T1- and hyperintense on T2-weighted MR images showing homogenous contrast enhancement. Our experience as well as of those in the literature show that total excision of these lesions is often possible with microsurgery and that the possibility of recurrence after local removal is extremely low.

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