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Dorsal Spinal Epidural Cavernous Haemangioma: A Case Report

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Abstract: *A 60 year old male patient presented with diffuse pain in the dorsal spine since 3 months with a previous history of having underwent a surgery for a spinal tumour 20 years ago, the details of biopsy of which was unavailable. On examination he had spastic paraparesis with no sphincter disturbances. Magnetic resonance imaging revealed an extramedullary space occupying lesion on the left side extending from D5 to D8 vertebrae causing severe cord compression with evidence of previous laminectomy from D9 to D11 levels. Radiological differential diagnosis included a nerve sheath tumour or a meningioma. He underwent D4-D8 laminectomy and intraoperatively the tumour was purely extradural with mild extension into the left neural foramen with no attachment to the nerves or the dura. Total excision of the extradural compressing mass was possible as there were preserved planes all around. Histopathology revealed cavernous hemangioma. .Though uncommon, purelyepidural hemangiomas must to be considered in the differential diagnosis of a spinal epidural soft tissue mass. Findings that help to differentiate this lesion fromdisk prolapse, meningiomas and nerve sheath tumors are its ovoid shape, uniform T2 hyperintense signal and lack of anatomic connection with the neighbouring intervertebral disk or the exiting nerve root. Complete extradural lesions without bony involvement are rare representing only about 12% of all intraspinalhemangiomas.*

Keywords:*Spinal, extradural, haemagioma.*

I. Introduction

The aim of the present study is to report an uncommon etiology for spinal extradural space occupying lesions, review the literature, thus emphasising the diagnostic importance of this lesion. In the present study, the differential diagnosis of extra-axial space occupying lesions of the spine has been briefly reviewed, highlighting the comparison of the imaging features of this lesion with other more common pathologies at this site.

II. Case Report

A 60 year old gentleman presented with mild back pain localised to the mid dorsal region with progressive heaviness and weakness of both lower limbs since 3 months. There was no history of associated radiating pain over the chest and abdomen and there was no evidence of sphincter disturbances. There was previous history of having underwent surgery for a spinal tumour 20 years ago, the details of which were unavailable. On examination, patient had spastic paraparesis with a power of 3/5 in both lower limbs and there was no local tenderness of the spine.

A plain magnetic resonance imaging (MRI) study of the dorsal spine showed an extramedullaryspace occupying lesion on the left side of the spinal canal from D5 to D8 vertebral levels. The mass was well marginatedwith mild extension into the adjacent neural foramen but there was no bone involvement. The lesion was hypointense on T1-weighted images [Figures 1& 3] and hyperintense on T2-weighted images [Figures 2 & 4], in relation to the intervertebral disk with severe compression and displacement of the adjacent thecal sac and spinal cord to the right side.Extension of the lesion into the left neural foramen was observed at D6 level. Radiological differential diagnosis included nerve sheath tumor and meningioma.

The patient underwent D4-8 laminectomy under general anesthesia. Intraoperatively, the tumor was purely extradural in location with mild extension into the left neural foramen. No attachment to the nerves or dura was found. The mass was predominantlygreyish red, soft with moderate vascularity noted than usual for a meningioma or a schwannoma. Lesionwas purely extradural with no intramedullary extension and haemostasis was achieved effortlessly. Hence, a total excision of the lesion was possible, without the need for postoperative adjuvant radiotherapy.

On histology, the lesion was composed of multiple, closely spaced, dilated and ectatic vascular channels containing blood. The spaces were lined by a single layer of benign endothelial cells. Nuclear pleomorphism and mitotic figures were not seen[Figures 5,6 and 7].In view of the above findings the final histopathology was reported as a cavernous hemangioma.

Patient showed remarkable improvement postoperatively. During the first follow up at the end of 4 weeks, the spasticity in the lower limbs disappeared and power in both lower limbs improved to 4+/5 with no fresh deficits.

III. Discussion

Cavernous hemangiomas were first reported in 1929 by Globus and Doshay, and are defined as benign vascular structures developed between the neural tissues occurring in the central nervous system, consisting of a dilated vascular bed [1]. They are congenital vascular malformations of unknown origin and can be clearly differentiated from true vascular neoplasms, such as hemangioblastomas. Cavernous haemangiomas grow slowly probably because of recurrent haemorrhage and thrombosis with organization and recanalization, in spite of absence of any mitotic activity. Hence they do not resolve spontaneously, requiring surgical intervention.

Cavernous angiomas or cavernomas can be sporadic or familial, out of which 10% are familial and the rest being sporadic[2]. They can occur anywhere in the central nervous system. About 80% of the cavernous angiomas occur supratentorially, 15% infratentorially and 5% are localized to the spine[3]. They constitute about 7% of all vascular malformations, and spinal vascular malformations account for about 3-12% of spinal space occupying lesions[3,4]. They can be solitary or multiple and may be associated with similar lesions at other sites in the body. In the spine, these lesions may be confined to the vertebrae, or such vertebral lesions may secondarily extend into the spinal canal, or may be purely epidural or purely intramedullary in location [4].

Purely extradural benign vascular lesions of the spine are rare and comprise less than 6% of all spinal neoplasms[5].Spinal cavernous hemangiomas most often originate from the vertebral bodies, sometimes with secondary extension into the extradural space [6]. Entirely extradural lesions with no bone involvement represent about 12% of all intraspinalhemangiomas[7,8,9].

Irrespective of the location, all cavernous haemangiomas share the same features on light and electron microscopy aswell as in immunohistochemistry[10]. They are composed of sinusoidal vessels situated adjacent to oneanother without intervening parent tissue. Grossly, they appeardark purplish in color, and arewell-circumscribed lesions with discrete borders, but can be multilobulated. There may be occasional calcification and thrombosis. They are angiographically occult as the blood flow through theselesions is slow and thus have no communication with the spinal vasculature[11].Intraaxial lesions tend to present more often with hemorrhagesthan the extraaxialones[12].

Clinically, patients with spinal cavernous haemangiomas present withfeatures suggestive of slowlyprogressive spinal cord compression. Rarely, theremay be a short history with acute compressive features due to eitherextradural haemorrhage or thrombotic venous occlusion [13]. The increased propensity to bleed can be explained by the thin-walled vessels and by stasis of blood flow in the lesion[14]. Other causes of acute symptoms include estrogen mediated neoangiogenesis in the lesion or direct compression by a gravid uterus.Our patient had short duration of symptoms with progressive spastic paraparesis, clinically suggesting a dorsal compressive myelopathy.

Computed tomographyscan may show a hyperdensecalcified lesion in long standing cases. There may be minimal or no enhancementwith contrast. MRI is the investigation of choice and may appear as an isointense lesion on T1 and iso to hyperintense in T2 weighted sequences. Contrast enhancement may be often seen. Chances of intratumoural bleed are rare and are mostly seen in lesions which appear hyperintense on T1 weighted imaging [15].On the other hand, the MRI features of cavernous angiomas of the brain and spinal cord is vastly different. In the brain, there arelow signal on T1 and T2 sequences correlating to repeated episodes of bleeding with hemosiderin deposition. The absence of a low-signal hemosiderin ring on both T1 and T2-weighted images in epidural lesions is likely to be related to more rapid removal of blood degradation products outside the blood-brain barrier[12.13,15]. Nerve sheath tumours, meningiomasand metastasis are the usual differential diagnoses for these extradural spinal lesions.

Microsurgical excision of these lesions in surgically accessible locations is the treatment of choice and radiotherapy is reserved only in cases where the lesions are excised incompletely either due to uncontrollable bleeding or those with intramedullary extension [16]. Patients recover completely after total excision of these tumours with no evidence of recurrent or residual lesions as reported in most of the previously reported cases [17,18,19,20]. In our case, lesion was purely extradural with no intramedullary extension and bleeding waseasily controlled. Hence, a total excision of the lesion was done, with no need for postoperative adjuvant radiotherapy.

IV. Conclusion

As suggested by the review of literature, the diagnosis of a spinal epidural cavernous hamangiomais usually missed on preoperative imaging, as encountered in our case. Hence, a high index of suspicion is necessary for the diagnosis based on preoperative imaging, and though the lesion is rare,cavernous hemangioma should be included in the differential diagnosis of purely extradural soft tissue lesions of the spine as early and accurate diagnosis followed by complete excision of the lesion before any possible episodes of massive intralesional bleeding is related to a favorable outcome.

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