Case Report

Epidural extension of dorsal vertebral D1–D7 hemangiomas with congenital cutaneous hemangiomas: Case report and literature review

Suresh S. Pillai1, P. A. Ramsheela2, Rijil Deepak3, Shinto Francis4, C. Jayakrishnan5, Ani Praveen6, Neena Mampally6

1Department of Spine Surgery, Baby Memorial Hospital, Calicut, Kerala, India, Departments of 2Orthopedics, Baby Memorial Hospital, Calicut, Kerala, India, 3Anaesthesia, Baby Memorial Hospital, Calicut, Kerala, India, 4Hemato-Oncology, Baby Memorial Hospital, Calicut, Kerala, India, 5Neurology, Baby Memorial Hospital, Calicut, Kerala, India, 6Pathology, Baby Memorial Hospital, Calicut, Kerala, India.

E-mail: *Suresh S. Pillai - sureshorth@gmail.com; P. A. Ramsheela - ramshi0046@gmail.com; Rijil Deepak - rjjdeepak69@gmail.com; Shinto Francis - meshinto@gmail.com; C. Jayakrishnan - jjkkecheri@gmail.com; Ani Praveen - ani.praveenkk@gmail.com; Neena Mampally - neenamampilly@gmail.com

ABSTRACT

Background: There are only rare reports of simultaneous multiple thoracic vertebral, epidural, and congenital cutaneous hemangiomas occurring at the same levels.

Case Description: A 24-year-old male presented with a progressive paraparesis attributed to multiple vertebral hemangiomas (MVH) with epidural extension (i.e. resulting in D1–D3 significant cord compression.), plus congenital cutaneous lesions at the D2–D7 levels. Following preoperative angioembolisation, a D1–D7 laminectomy was performed along with a C7–D8 pedicle screw fixation. Pathologically the bone and cutaneous lesions were spinal cavernous hemangiomas. Postoperatively, the patient regained normal function. As complete excision was not feasible, he subsequently received radiotherapy to prevent tumor recurrence.

Conclusion: MVH with multilevel epidural extension resulting in significant cord compression and congenital cutaneous lesions should undergo attempted tumor excision followed by radiation therapy where complete removal is not feasible.

Keywords: Compressive myelopathy, Congenital cutaneous hemangioma, Epidural hemangioma, Vertebral hemangioma

INTRODUCTION

Multiple vertebral hemangiomas (MVH) typically involve multiple vertebral bodies, and occasionally extend into the epidural space. They represent 2–3% of all spinal tumors, and usually follow an indolent, asymptomatic course. However, a subset may develop symptoms/signs of significant cord compression, attributed to combinations of epidural cord compression, pathological fractures, and/or hemorrhages, warranting surgery. Histologically, MVH are characterized into one of three categories; the capillary, cavernous, arteriovenous, or the venous type. Here, a 24-year-old presented with the new onset of a paraparesis attributed to MVH extending from D1 to D7 with epidural extension/cord compression and accompanying cutaneous lesions at the same levels. The spinal lesion required extensive...
thoracic cord decompression with fusion, followed by radiation therapy to address residual disease.

**CASE REPORT**

A 24-year-old male presented with 6 months of a progressive paraparesis that exacerbated over the past 2 months. Notably, he had a large cherry red cutaneous lesion on the left side of the thorax, (i.e. left side of the chest from sternum to mid line at the back), also involving the left arm/forearm (i.e. from the nape of neck to just below the scapula involving the lateral/medial arm/forearm) since birth [Figures 1a and b].

**Neurological**

On examination, he had 3–4/5 motor strength diffusely in the lower extremities and a relative sensory level below D4 level without sphincter involvement.

**CT Findings**

The thoracic CT showed irregular lytic lesions with coarse thickened trabeculae involving the D1 through D7 vertebral levels, also including the left 1st through 6th ribs. Axial studies showed the typical “polka-dot” pattern of vertebral involvement [Figure 2]. There was also left sided paraspinal and intracanalicular enhancing soft tissue thickening adjacent to the involved vertebrae resulting in significant cord compression.

**MR findings**

On the MR, tumor was hypointense on both T1W and T2W images and enhanced with contrast (i.e., encasing the cervicodorsal spinal cord from C7 to D8 with transforaminal extension). There was also a subtle increased intracord T2W hyperintensities seen at the D2-D3 vertebral levels [Figures 3-5].

**Thoracic CT angiography (CTA) with embolization**

The Thoracic CTA showed a highly vascular lesion extending from D1 to D7 that was mostly left-sided (i.e. extra and intracanalicular tumor), the major feeders were embolized before surgery (i.e., with gel foam and coils).

**Surgery**

Following a D1–D7 laminectomy, a red brown, soft lesion was encountered a leash of blood vessels. Tumor was adherent to the dorsal thecal sac and demonstrated lateral, foraminal, and left-sided paraspinal extension [Figure 6a]. The blood vessels were coagulated and removed [Figure 6b]. Finally, the
Patient underwent a pedicle screw fusion from C7 to D5–D8 [Figure 6c and Figure 7].

Pathology

The frozen sections (i.e., from the epidural, lamina, paraspinal, and skin) were all consistent with a cavernous hemangioma [Figures 8a-c and Table 1].

Postoperative recovery

The patient had an uneventful recovery; 1 week later, at discharge, he had improved and was able to ambulate without weakness. Two months later his muscle strength was 5/5 bilaterally.

Postoperative adjunctive radiation therapy

The patient underwent postoperative radiotherapy to the tumor bed to treat surgically inaccessible tumor, and limit the chance of recurrence.
DISCUSSION

According to the WHO classification, vertebral hemangiomas are considered to be a benign vasoformative neoplasms of bone. The most common site is the thoracic (approximately 60%), followed by cervical (30%) and lumbar (10%) spine. These lesions are usually confined to the vertebral bodies, but occasionally may extend into the pedicles, arches, and spinous processes. Notably, the vast majority of vertebral hemangioma are asymptomatic, are usually diagnosed incidentally on radiological imaging or during postmortem studies. The further epidural extension of vertebral hemangioma is seen only in 1–2% of cases, contributing to slowly progressive compressive myelopathies or radiculopathies.

In the case we presented, the patient has a congenital cutaneous hemangioma at same levels as the vertebral/epidural tumor sites. Whereas, some literature describes case with concomitant occurrence of haemangiomas in other organs like liver and spleen in association with spinal haemangioma. MR imaging is the most reliable investigation for spinal hemangioma. Spinal angiography is recommended prior to surgery and preoperative angiobolization of feeding vessels should be done, to avoid the risk of massive bleeding intraoperatively. Laminctomy is an effective procedure to decompress the cord without contributing to instability or deformity. If, however, vertebral body compression fractures occur, vertebroplasties and/or fusions may be warranted.

CONCLUSION

MVH with epidural extension plus congenital cutaneous lesions rarely occur simultaneously at the same levels. In the presence of compression, gross total excision of tumor is optimal, but subtotal removals may be followed by adjunctive radiation therapy to limit the chance of tumor recurrence.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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