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Pregnancy and Aggressive Angioma: A Very Rare Entity: Case Report and Literature Review

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Abstract **Case Report**

Vertebral hemangiomas (VH) are common benign tumors, often asymptomatic. Their clinical presentation during pregnancy is rare. We report the case of a 38-year-old woman at 37 weeks of pregnancy, admitted for progressive paraplegia caused by an aggressive VH at the D5 level. MRI revealed spinal cord compression, necessitating a cesarean section followed by laminectomy and cementoplasty. The growth of VH during pregnancy is facilitated by hemodynamic and hormonal changes. Treatment involves surgery, with embolization as an option to reduce bleeding, and radiotherapy in the postpartum period. This rare case highlights the importance of early diagnosis and multidisciplinary management.

Keywords: Vertebral hemangioma, pregnancy, spinal cord compression, laminectomy.

INTRODUCTION

Vertebral hemangiomas are benign, slow-growing tumors. They are commonly observed in females and are predominantly located in the thoracic spine, representing 10 to 12% of spinal tumors. Symptomatic cases account for only 1% of occurrences and are typically discovered incidentally. The association between a symptomatic vertebral hemangioma and pregnancy is of particular interest due to the potential correlation with the physiological changes that occur during pregnancy, especially the hemodynamic and endocrine alterations in the third trimester, which may play a pivotal role in the expansion and enlargement of hemangiomas [1-3].

We present a new case of vertebral compression due to hemangioma during the third trimester of pregnancy. Through this case and a comprehensive review of the literature, we aim to emphasize the diagnostic modalities, therapeutic management, and prognosis of this rare pathology.

CASE REPORT

A 38-year-old female, G5P4, with no significant medical or surgical history, was admitted to the Obstetrics Department for flaccid paraplegia with sensory loss below the umbilical level at 37 weeks of gestation. The patient delayed seeking medical consultation until 20 days after the onset of her initial symptoms.

The first symptom was distal weakness in the left lower limb, accompanied by edema, which was followed by weakness in the right lower limb a week later. Ten days prior to admission, the patient developed complete weakness in both lower limbs, associated with lumbar pain, and acute urinary retention for one day. There were no symptoms in the upper limbs, nor any fever, headaches, or vomiting.

Neurological examination upon admission revealed segmental muscle strength graded 0/5 in the lower limbs and 5/5 in the upper limbs, flaccid tone in the lower limbs, and preserved knee reflexes but absent Achilles reflexes bilaterally. The patient also exhibited hypoesthesia in the lower limbs, with a tactile and pain sensory level located above the umbilicus.

Obstetric evaluation, including clinical, ultrasound, and cardiotocographic assessments, confirmed normal fetal status with regular movements. The patient's laboratory results were unremarkable.

Magnetic resonance imaging (MRI) revealed a signal abnormality centered on the D5 vertebral body,

extending to the posterior arch and exhibiting a "salt-and-pepper" appearance. The posterior cortical bone was breached, with an intra-canal extension and mass effect on the spinal cord, which appeared swollen and exhibited T2 hyperintensity, consistent with an aggressive hemangioma of the D5 vertebral body causing spinal cord compression with signs of cord distress (Figures 1 and 2).

A cesarean section was performed, followed on the same day by a laminectomy of the 4th and 5th dorsal vertebrae, along with cementoplasty of D4.

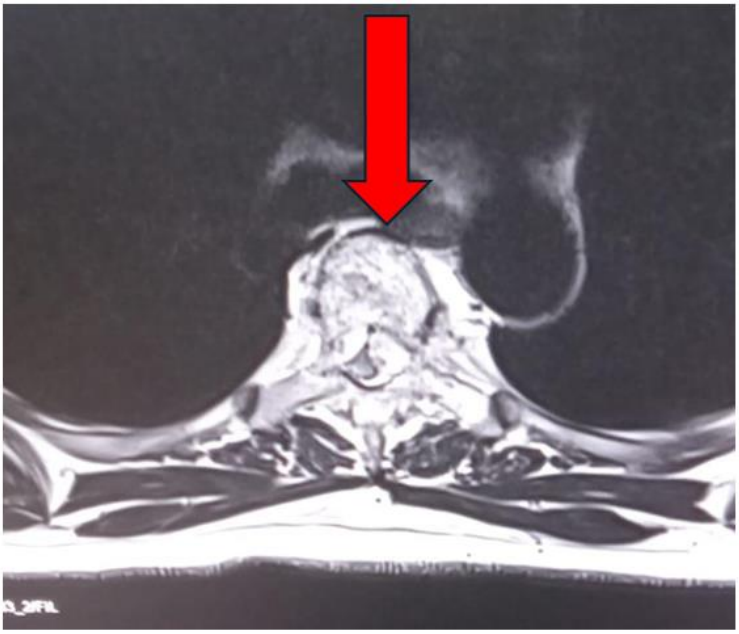

Fig 1: Showing the involvement of the vertebral body (VB)

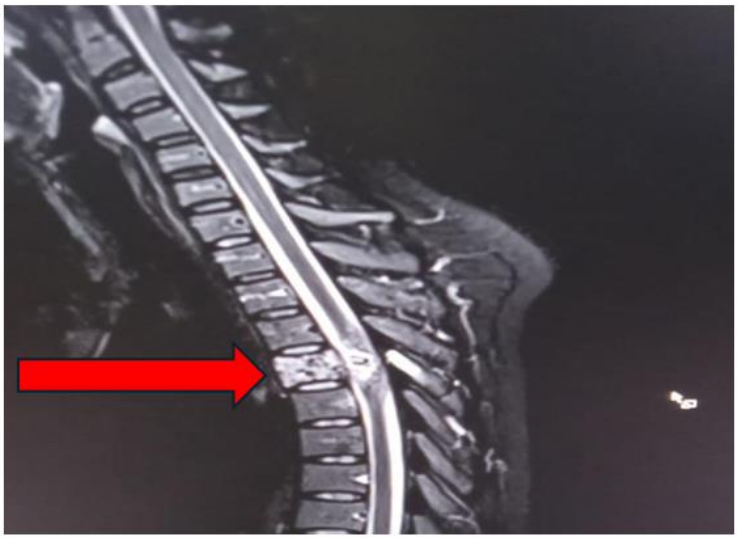

Fig 2: Sagittal view showing the level of the lesion and the mass effect on the spinal cord

DISCUSSION

Most vertebral hemangiomas (VH) are asymptomatic, with the thoracic spine being the most commonly affected region. VH can cause neurological symptoms through various mechanisms, including the expansion of bony elements, epidural expansion of tumor tissue, disruption of local blood flow, or, more rarely, compression fractures of the vertebral body associated with epidural hematomas [4, 5].

The first case of symptomatic vertebral hemangioma associated with pregnancy was described by Guthkelch in 1948 [4]. Since then, more than 30 cases have been reported in the literature, mainly in the third trimester, with only one case reported in the second trimester.

Several hypotheses have been proposed to explain the presentation of vertebral hemangiomas in pregnant women. Some authors suggest that hemangiomas may become more invasive and increase

in size during pregnancy due to a decrease in bone density, though this theory is not widely accepted. Two main factors are considered to contribute to hemangioma expansion:

1. **Hemodynamic**: There is up to a 50% increase in blood volume during pregnancy and increased venous pressure caused by compression of the inferior vena cava by the gravid uterus. This raises intra-abdominal pressure, leading to a redistribution of blood flow, particularly through the vertebral venous plexus, which increases the volume of the hemangioma and its associated mass effect [1, 6].
2. **Hormonal**: Elevated levels of progesterone and estrogen during the third trimester promote endothelial proliferation and venous distensibility due to increased plasma volume. Relaxin, a hormone produced by the placenta, may also play a role through its vasodilatory effect [1, 3, 6].

MRI is the gold standard for diagnosing vertebral hemangiomas, owing to its superior soft-tissue resolution and ability to finely assess the spinal cord. Additionally, the absence of ionizing radiation is a significant advantage given the fetus's sensitivity to radiation. Typical hemangiomas show hyperintensity on both T1- and T2-weighted sequences, reflecting the presence of intralesional fat, blood vessels, and interstitial edema [1, 7].

Aggressive hemangiomas, so named due to their rapid expansion leading to spinal cord or nerve compression, contain less fat and more vascular stroma than non-aggressive hemangiomas, and thus present with decreased signal intensity on T1-weighted sequences [7, 8]. Key radiological features of aggressiveness include cortical erosion, epidural soft tissue expansion from posterior elements, and invasion of the spinal canal, as reported in our case.

In terms of treatment, laminectomy is the standard surgical technique for these tumors, which carry a significant risk of extensive bleeding. Selective arteriography and embolization are useful in neurologically stable patients. Embolization reduces intraoperative bleeding and may, in some cases, be sufficient as the sole treatment for the hemangioma. Radiotherapy, although contraindicated during pregnancy, is sometimes necessary as an adjunct to surgery in the postpartum period [9-11]. Postpartum radiotherapy was administered to our patient following laminectomy with good outcomes.

There is no consensus on the optimal timing of surgery, but factors such as gestational age, fetal maturity, and the severity of the mother's neurological deficits must be carefully considered in a multidisciplinary discussion.

CONCLUSION

Pregnant patients with symptomatic vertebral hemangiomas represent a rare phenomenon. The diagnosis of spinal cord compression may be delayed due to various clinical factors that could divert attention. It is crucial to consider the possibility of spinal cord compression related to a pre-existing hemangioma in a previously healthy pregnant woman presenting with lower limb neurological deficits. Optimized multidisciplinary management undoubtedly contributes to an improved prognosis.

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