

## Case Report

# Aggressive vertebral hemangioma in the postpartum period: an eye-opener

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Pregnancy is a well-known risk factor for incidental or asymptomatic vertebral hemangiomas becoming aggressive or symptomatic, most often during the third trimester of pregnancy, related to hemodynamic and endocrinal changes occurring during pregnancy. Many patients show spontaneous incomplete remission after delivery. We report a rare case of aggressive vertebral hemangioma in the postpartum period in a 26-year-old woman, who presented with upper backache with progressive spastic paraparesis.

## INTRODUCTION

Vertebral hemangiomas are benign vascular tumors in the spinal column, present in 10–12% of all patients, more common in females [1, 2]. Symptomatic vertebral hemangiomas are rare and represent <1% of all hemangiomas. These lesions are solitary in 66% and multiple in 34% of cases. The majority of vertebral hemangiomas occur in thoracic spine, whereas lumbar and cervical spine is less favored sites. Most of the symptomatic vertebral hemangiomas are located in the thoracic region, specifically between T3 and T9. Pregnancy is a well-recognized state during which asymptomatic vertebral hemangiomas become symptomatic, most often in the third trimester of pregnancy due to physiological changes occurring during pregnancy. Many patients show spontaneous improvement after delivery [3].

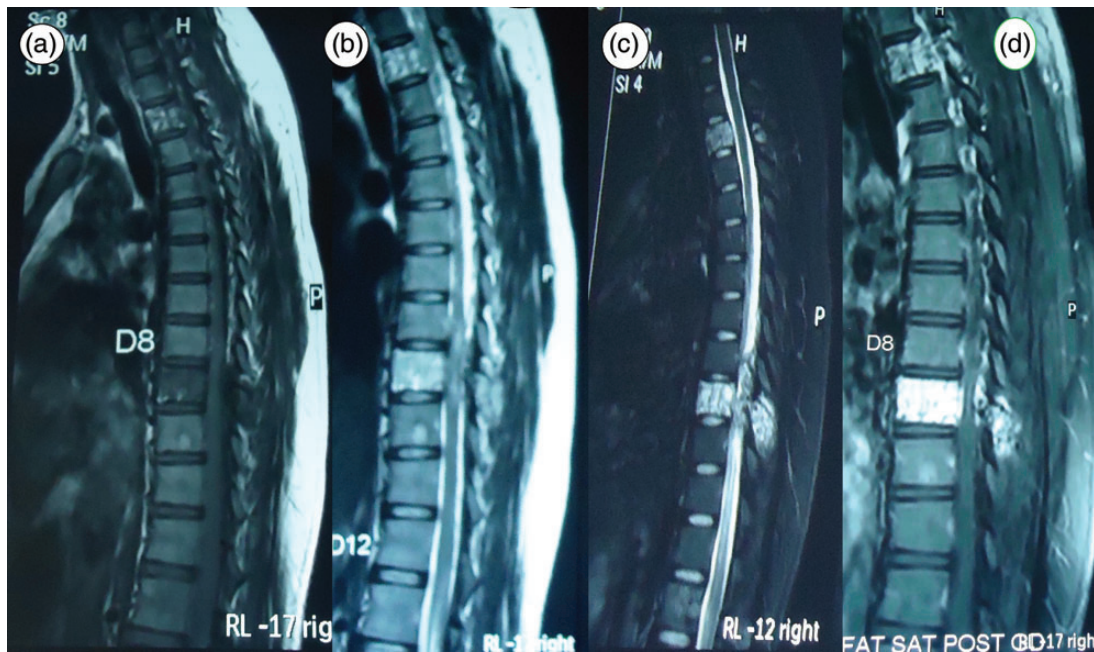
This report describes a rare case of aggressive vertebral hemangioma in the thoracic region in the postpartum period causing compressive myelopathy secondary to epidural extension.

## CASE REPORT

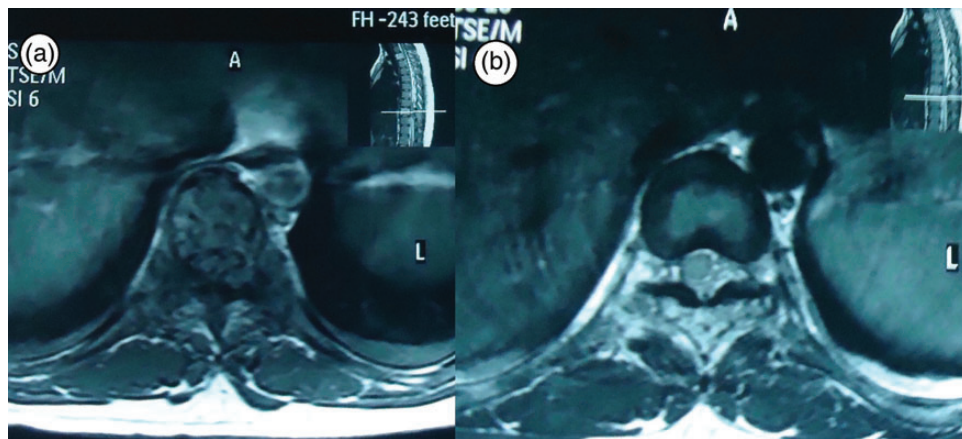
A 26-year-old postpartum female presented with 1-month history of backache with pain radiating to both the lower limbs, numbness in legs and progressive difficulty in walking. She became bed ridden within 15 days of onset of symptoms. There was no history of fever, headache, constitutional and

bowel/bladder symptoms. Her pregnancy period was unremarkable and she had a normal delivery. The general physical examination and examination of other systems (Gastrointestinal Tract, Cardiovascular System and respiratory) were normal. Neurological examination revealed spasticity in both the lower limbs with motor power Medical Research Council grade 1/5. Knee and ankle reflexes were exaggerated bilaterally with positive Babinski's sign. Sensory level was noted at T11 spinal level. The examination of upper limbs was unremarkable. There were no cerebellar signs.

The magnetic resonance imaging (MRI) revealed heterogeneous lesion (altered marrow signal intensity) diffusely involving body and posterior elements of D9 vertebra. The lesion appeared heterogeneously hyperintense on  $T_1$ -weighted image (Fig. 1a), hyperintense on  $T_2$ -weighted image (Fig. 1b) and markedly hyperintense on short  $T_1$ -inversion recovery (STIR) image (Fig. 1c). Extrasosseous extension of the lesion was seen in paravertebral region bilaterally, and showed similar signal changes. Epidural soft tissue component was also seen encasing the cord all around and compressing it. Cortical erosions were also seen in the affected vertebrae. Multiple areas of signal voids were seen within the lesion, likely representing the thickened trabeculae and flow voids (vessels; Fig. 2a and b). The lesion showed intense enhancement on post-contrast scan (Fig. 1d). Similar lesion was also seen in D2 vertebra showing subtle cortical erosion with no evidence of cord compression at this level. Spinal



**Figure 1:** Sagittal  $T_1$ -W,  $T_2$ -W, STIR and post-contrast MR images of the spine showing heterogeneous lesions (altered marrow signal intensity) involving body and posterior element of D9 vertebra, which is heterogeneously hyperintense on  $T_1$ -W (a), hyperintense on  $T_2$ -W (b), markedly hyperintense on STIR (c) and showing intense contrast enhancement on post-contrast image (d). Altered marrow signal intensity is also seen in D2 vertebra.



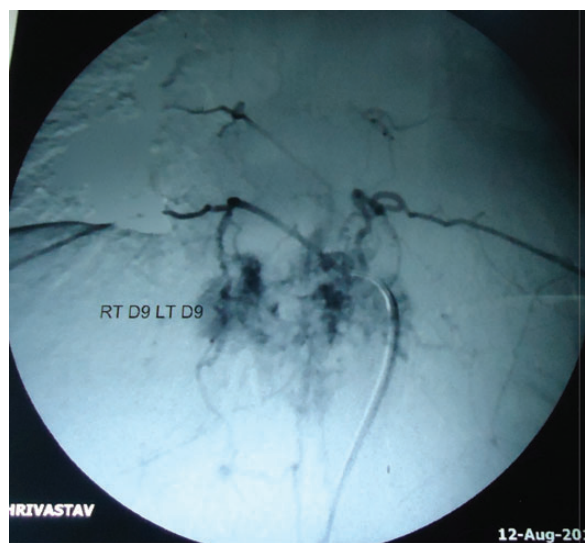
**Figure 2:** Axial  $T_1$ -W and  $T_2$ -W MR images of the spine (a and b) showing extraosseous extension of the lesion with epidural soft tissue component, encasing and compressing the cord. Multiple areas of signal voids are also seen. Affected vertebrae shows cortical erosion as well.

angiography was advised to look for the vascularity of the lesion and showed diffuse blush at the site of lesion (Fig. 3). There was no well-formed nidus or prominent vein to suggest arteriovenous malformation. Metastasis of the spine was considered but excluded because she was too young, there were no constitutional symptoms and the presence of hyperintense signals on  $T_1$ -weighted image favors benign etiology. Also, the vertebral height was preserved despite multiple lesions, again suggesting benign pathology. Metastasis of the spine usually shows destruction of the trabeculae and variable contrast enhancement. Hence, aggressive hemangioma of the thoracic spine in the postpartum period was considered, based on the erosion of vertebral bodies and extraosseous extension of the lesion. Patient was advised for preoperative

embolization followed by surgical excision of the tumor, but she refused for the surgery and lost to follow-up; hence, final outcome of the patient without surgery and histopathological confirmation was not possible to ascertain.

## DISCUSSION

Although normally being referred to as a benign tumor, vertebral hemangioma is essentially a vascular malformation. The first case of pregnancy-related symptomatic vertebral hemangioma was described in the year 1927 by Balado [3]. Most of them usually present during the third trimester and may show improvement in the postpartum period. Vertebral hemangiomas tend to have recurrence in subsequent pregnancy



**Figure 3:** Spinal angiography showing diffuse blush at the site of lesion.

with increased severity of myelopathic symptoms. The physiological factors including vascular, hormonal and altered hemodynamics act in concert to enlarge the pre-existing vertebral hemangioma. Venous obstruction and increased intra-abdominal pressure caused by gravid uterus cause redistribution and increased blood flow through vertebral venous plexus, leading to growth and expansion of pre-existing vertebral hemangiomas [4, 5]. An increase in plasma volume, increased venous distensibility due to high levels of maternal progesterone and the endothelial growth promoting effects of estrogen, seen in pregnancy, are other factors favoring growth of vertebral hemangioma [6]. In vertebral hemangioma, cord compression may be produced by one or more of the following mechanisms: (i) narrowing of spinal canal by the expansion of tumor within the vertebral body and/or posterior elements, (ii) subperiosteal growth of hemangioma forming an epidural mass, (iii) acute hemorrhage into the epidural space and (iv) rarely, compression fracture of the involved vertebral body [7]. Compression fracture in hemangioma is unusual because the involved vertebra usually have thickened vertical trabeculae, which can withstand axial collapse. Vertebral hemangioma typically shows a coarse trabecular pattern, described as a 'jail bar' or 'corduroy' appearance on plain radiographic films [8]. On axial computed tomography, this coarse trabecular pattern is characteristically seen as dense spots within a hypodense lesion, referred to as a 'polka dot' pattern [9]. On MRI, benign, incidental hemangiomas usually appear as hyperintense on both  $T_1$ - and  $T_2$ -weighted images due to high fat component and less vascular stroma. On the other hand, aggressive hemangiomas are usually hypointense to isointense on  $T_1$ -weighted images and hyperintense on  $T_2$ -weighted images due to prominent hypervascular stroma and less amount of fat [10]. These lesions show marked contrast enhancement in contrast to spinal metastases, which shows variable enhancement [11]. MRI is also helpful in showing

paravertebral and epidural extension of the lesion. Extraosseous components lack fatty tissue and thus appear isointense on  $T_1$ -weighted images.

The third trimester of pregnancy has been reported as the most favorable time for symptomatic presentation of vertebral hemangioma, but presentation in the postpartum period has also been reported rarely [12].

Treatment of vertebral hemangiomas is indicated if progressive neurological deficit or severe pain develops. Treatment options include radiation therapy, embolization, percutaneous sclerotherapy, vertebroplasty and/or surgical decompression.

Till date, compression fracture of the involved vertebral body was reported as a rare cause in a case of symptomatic vertebral hemangioma presenting in the postpartum period [12]. To the best of our knowledge, this is probably the first case of aggressive vertebral hemangioma causing progressive spastic paraparesis in the postpartum period due to subperiosteal extension of hemangioma, resulting in an extradural mass and thereby compressing the spinal cord. The occurrence of compressive myelopathy in the postpartum period is quite unusual and could represent the perturbed hemodynamics milieu resulting in expansion of the pre-existing vertebral hemangioma. This case report should be an eye-opener for clinicians, meaning that vertebral hemangioma can manifest aggressively not only during the third trimester of pregnancy but also in the postpartum period.

## CONFLICT OF INTEREST STATEMENT

None declared.

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