

Case Report

Recurrent symptomatic vertebral hemangioma during two consecutive pregnancies: Case report and review of the literature

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Abstract

Background: Pregnancy-related changes can exacerbate the symptoms/signs of vertebral hemangiomas. Here, we report a patient who experienced symptomatic vertebral hemangiomas resulting in cord compression during two consecutive pregnancies.

Case Description: A 28-year-old female 34 weeks pregnant, presented with a progressive spastic paraparesis. Magnetic Resonance Imaging (MRI) demonstrated an T5 vertebral body signal change attributed to a hemangioma resulting in cord compression. Following a cesarean section, she had a trans thoracic T5 corpectomy with spinal fusion. Indeed, the histopathology was consistent with a vertebral hemangioma. She fully recovered after this first surgery. However, six years later, she again presented with a spastic paraparesis and sphincter deficit now 29 weeks pregnant. The MR demonstrated cord compression one more at the T5 level attributed to the hemangioma; following a T5 and T6 laminectomy, the left paracentral epidural vascular mass totally resected. Her child was successfully delivered 2 months later at which point she exhibited only mild residual lower limb spasticity.

Conclusion: Patients with known vertebral hemangiomas should be closely monitored during pregnancy as increased growth during these pregnancies may result in progressive spinal cord compression.

Key Words: Pregnancy, symptomatic hemangioma, tumor recurrence, vertebral hemangioma

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INTRODUCTION

Pregnancy can exacerbate the growth/presentation of vertebral hemangiomas resulting in increased epidural spinal cord compression. Here, the authors report one patient with two episodes of paraparesis attributed to a T5 hemangioma during two separate pregnancies. The literature concerning 37 other comparable cases of hemangiomas becoming symptomatic during pregnancy were also reviewed.

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CASE DESCRIPTION

A 28-year-old female in her 34th week of pregnancy presented with progressive paraparesis over a 10-day period. On examination, she was paraplegic with a sensory level and hyperreflexia. The magnetic resonance imaging (MRI) demonstrated a T5 vertebral body hemangioma characterized by an isointense signal on T1 and hyperintense signal on T2-weighted images, which contributed to marked cord compression. After cesarean section, she underwent a transthoracic T5 corpectomy, T4/5 and T5/6 discectomy, and excision of an epidural vascular mass. This was completed by spinal fusion using an expandable cage, screw/rod construct, and bone graft [Figure 1]. Histopathologically, this proved to be a cavernous vertebral hemangioma. The patient fully recovered 3 months later. Six years later, however, she again presented with acute paraparesis/sphincter deficit, in the 29th week of her next pregnancy. The MRI demonstrated cord compression at the level of T5 attributed to hemangioma [Figure 2]. She underwent a T5 and T6 laminectomy with left paracentral excision of an epidural vascular mass. Postoperative, she had mild residual spasticity.

DISCUSSION

Vertebral hemangioma is the most common benign spinal neoplasm that occurs in 10–12% of the general population.^[3] In majority of patients, it is asymptomatic and diagnosed incidentally, but in about 1% of people it becomes symptomatic.^[3,7] The most common site for involvement is the lower dorsal and lumbosacral spine, and it only rarely involves the cervical spine.^[3] Rarely, it causes cord compression due to vertebral expansion and bleeding to the epidural space, rare pathologic fractures, and/or ischemia due to vascular shunting.^[2,3,7] The two main mechanisms for exacerbation/dilatation/expansion

of hemangiomas during pregnancy include: (1) increased blood volume/and elevated venous pressure particularly after seven months (e.g., greater uterine volume and inferior vena cava compression), and (2) hormonal changes and the angiogenic effects of estrogen and progesterone promoting growth.^[6,9,11]

Literature summary

Thirty-seven cases of symptomatic vertebral hemangiomas during pregnancy have been reported [Table 1].

Kiroglu *et al.* (2009) noted 27 previous case reports about pregnancy-related symptomatic hemangioma in patients averaging 27.8 years of age. They were located in the thoracic spine (23: 17 above T8), cervical spine (1 patient), and the lumbar spine (2 patients).^[4]

Moles *et al.* (2014) reported 27 cases of pregnancy-related symptomatic hemangiomas; 12 presented at less than 32 weeks, 8 between 32 to 36 weeks, and 7 > 36 weeks. They averaged 30 years of age, and lesions were located mostly in the thoracic spine (89%: 63% in the upper thoracic region).^[7] All patients presented with increased neurological deficits attributed to vertebral bone expansion or epidural vascular compression.^[7]

Several other studies reported comparable instances of hemangiomas resulting in acute paraparesis at different but latter stages of pregnancy.

Proposed management of vertebral hemangiomas in pregnancy

Chi *et al.* proposed an algorithm for managing vertebral hemangioma during pregnancy: if the patient is neurologically intact, the management is conservative despite the gestational age. But if the patient has a significant neurological deficit, and the gestational age <32 weeks, antepartum surgery should be performed, whereas a gestational age of >32 weeks, the fetus should first be extracted, and surgery should follow.^[1]

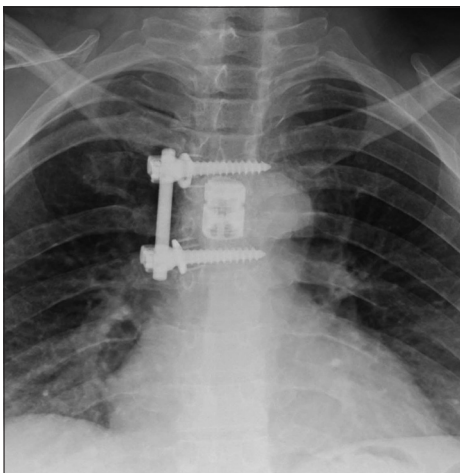


Figure 1: Postoperative X-Ray demonstrated T5 corpectomy and spinal fusion helping expandable cage, screw and rod

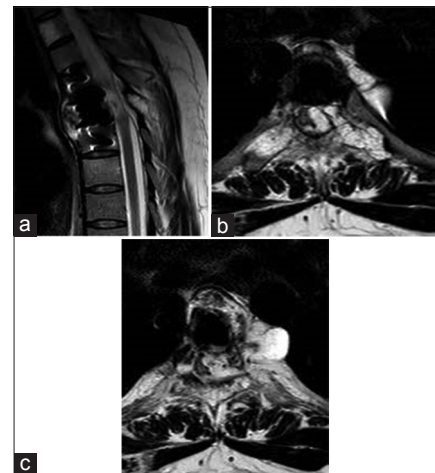


Figure 2: Demonstrated epidural mass compressing the cord. (a) The sagittal view (b and c) The axial view of spinal MRI

Table 1: Some of the most important features of spinal hemangioma during pregnancy

Study	Year	Study type	Patients number	Age	Level	Gestational age	Treatment
Kiroglu, <i>et al.</i> ^[4]	2009	Literature review	27	Mean: 27.8 years	Dorsal: 23 Cervical: 1 Lumbar: 2	6-9 months: 8 24<6 months: 3	Prepartum resection: 8 Postpartum resection: 16 Conservative: 1 Unknown: 2
Moles, <i>et al.</i> ^[7]	2014	Literature review	27	Mean: 30 years	Dorsal: 89% patients	<32 weeks: 12 32-36 weeks: 8 >36 weeks: 7	Prepartum resection: 8 Postpartum resection: 19
Ravikiran, <i>et al.</i> ^[8]	2013	Case report	1	25 years	T1/T2	26 weeks	Prepartum tumor embolization/resection
Slimani, <i>et al.</i> ^[10]	2014	Case report	1	19 years	T4	38 weeks	Postpartum resection
Gupta, <i>et al.</i> ^[3]	2014	Case report	1	23 years	T1-T5	32 weeks	Prepartum resection
Korkmaz, <i>et al.</i> ^[5]	2016	Case report	1	33 years	T8/T9	35 weeks	Postpartum resection
Our study	2017	Case report	1	First 28 years Recurrence 34 years	T5 T5	34 weeks 29 weeks	Postpartum resection Prepartum resection

CONCLUSION

It is important to monitor a patient with a vertebral hemangioma for the potential onset of a significant neurological deficit during pregnancy. Hemangiomas can demonstrate increased growth/expansion during pregnancy, and if a neurological deficit acute presents, patients will warrant emergency surgical decompression. Furthermore, despite attempted complete initial resections, recurrent pregnancies may result in recurrent symptomatic hemangiomas.

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Conflicts of interest

The authors declare that they have no conflict of interest, financial or otherwise with any organization.

REFERENCES

- Chi JH, Manley GT, Chou D. Pregnancy-related vertebral hemangioma. Case report, review of the literature, and management algorithm. *Neurosurg Focus* 2005;19:E7.
- Fox MW, Onofrio BM. The natural history and management of symptomatic and asymptomatic vertebral hemangiomas. *J Neurosurg* 1993;78:36-45.
- Gupta M, Nayak R, Singh H, Khwaja G, Chowdhury D. Pregnancy related symptomatic vertebral hemangioma. *Ann Indian Acad Neurol* 2014;17:120-2.
- Kiroglu Y, Benek B, Yagci B, Cirak B, Tahta K. Spinal cord compression caused by vertebral hemangioma being symptomatic during pregnancy. *Surg Neurol* 2009;71:487-92; discussion 92.
- Korkmaz M, Gok M, Cinar C, Guneyli S, Duzgun F, Oran I. Progressive spinal epidural hemangioma in pregnancy. *Spine J* 2016;16:e523-5.
- Lavi E, Jamieson DG, Granat M. Epidural haemangiomas during pregnancy. *J Neurol Neurosurg Psychiatry* 1986;49:709-12.
- Moles A, Hamel O, Perret C, Bord E, Robert R, Buffenoir K. Symptomatic vertebral hemangiomas during pregnancy. *J Neurosurg Spine* 2014;20:585-91.
- Ravikiran HG, Harish RP, Ravikumar M, Sandeep N, Roopesh RG, Patil RK. Symptomatic Capillary Hemangioma in Pregnancy: A Rare Case Report of Complete Recovery of Paraparesis with Review of Literature. *IJHSR* 2013;3:161-6.
- Schwartz TH, Hibshoosh H, Riedel CJ. Estrogen and progesterone receptor-negative T11 vertebral hemangioma presenting as a postpartum compression fracture: Case report and management. *Neurosurgery* 2000;46:218-21.
- Slimani O, Jayi S, Fdili Alaoui F, Bouguern H, Chaara H, Fikri G, *et al.* An aggressive vertebral hemangioma in pregnancy: A case report. *J Med Case Rep* 2014;8:207.
- Vijay K, Shetty AP, Rajasekaran S. Symptomatic vertebral hemangioma in pregnancy treated antepartum. A case report with review of literature. *Eur Spine J* 2008;17(Suppl 2):S299-303.