

Aggressive vertebral hemangioma presenting in postpartum period: A case report

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ABSTRACT

Pregnancy is a known risk factor for hemangiomas to become aggressive and symptomatic. This is mainly because of the hormonal and hemodynamic changes and also due to increased intra-abdominal pressure. Vertebral hemangiomas usually become symptomatic in the third trimester. We present a rare case in which a young 20-year-old woman who remained asymptomatic during her pregnancy and presented with progressive paraparesis 5 days after vaginal delivery. Magnetic resonance imaging was done which revealed a vertebral hemangioma also involving the posterior elements, with soft-tissue extension in posterior epidural space. Surgery was done with removal of the soft tissue which was purely extradural in location. Histopathology revealed cavernous hemangioma.

Key words: Aggressive, Postpartum period, Vertebral hemangioma

Vertebral hemangiomas are benign lesions of the vertebrae. They are vascular lesions which may sometimes present with neurologic symptoms. They can be locally aggressive with extra-vertebral extension. There is 10–12% incidence of vertebral hemangiomas [1]. They are symptomatic in only 1% of cases [2]. It is important to make a diagnosis of hemangioma preoperatively by magnetic resonance imaging (MRI) as hemorrhage during resection and failure to remove it completely can lead to the persistence of clinical symptoms.

Pregnancy is known to cause asymptomatic hemangiomas symptomatic. They usually present in the third trimester. This is a rare case as it is presented postpartum period and only two such cases have been reported to our knowledge. We describe the case of a 20-year-old woman who presented with paraparesis 5 days after delivery. She was asymptomatic during her 9 months of pregnancy.

CASE REPORT

A 20-year-old woman presented with sudden onset of paraparesis 5 days after vaginal delivery which worsened rapidly over the next week. There was bladder urgency for 3–4 days. At presentation, the pulse, temperature, respiratory rate, and blood pressure were normal. The neurologic examination demonstrated decreased strength with spasticity of bilateral lower limbs and decreased sensation below the T6 level. Per rectal examination revealed decreased anal tone.

The blood investigation such as complete blood count was normal. The liver and renal function test reports were

unremarkable. MRI was done in a 3 tesla (Magnetom) MRI scanner. Contrast-enhanced sequences were obtained using 10 mg of gadolinium injection. Multiplanar MRI of the dorsal spine was done. T1 and T2 spin-echo images in the sagittal, transverse, and coronal planes were obtained. Fat-saturated images and post-gadolinium T1FS images were also taken in all three planes. MRI revealed an enhancing T1 hypointense and T2 and STIR hyperintense focal lesion in the posterior aspect of the D5 vertebral body (Fig. 1) also involving the posterior elements characteristic of vertebral hemangioma. There was lentiform shaped enhancing soft-tissue extension into the epidural space causing anterior displacement of spinal cord and extension into the right neural foramina (Fig. 1). The spinal cord signal was normal.

Surgery was immediately done under general anesthesia. Laminectomy was done and the cord was decompressed by removal of the epidural soft tissue. Intraoperatively, the tumor was purely extradural in the location with mild extension into the right neural foramina. It was soft gray in color and vascular. The entire tumor was not removed, only the soft tissue indenting the spinal cord was removed (Fig. 2). The histological examination showed vessel proliferation with thin wall and dilated lumen, covered by flat endothelial cells without atypia or mitosis, consistent with a cavernous hemangioma (Fig. 3). Post-operative limited MRI T2 spin echo sagittal and transverse images were done (Fig. 4).

The patient was followed up after 1 year. She was completely symptom free. Non-contrast MRI was done in the same machine which revealed a small residual hemangioma.

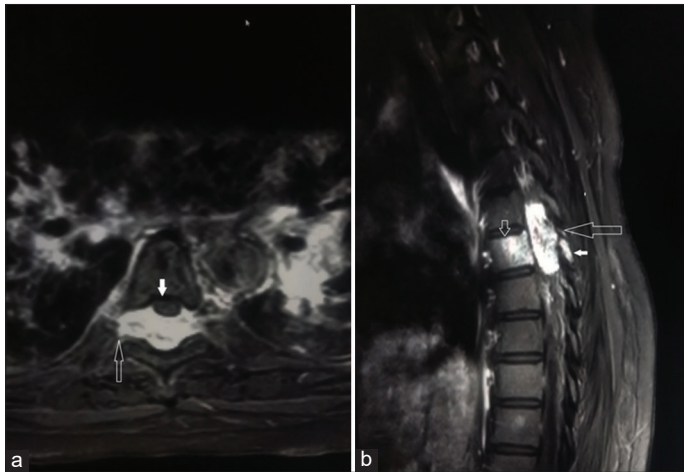


Figure 1: (a) Axial post-contrast T1 fat-suppressed image at D4 level showing brilliantly enhancing mass in the posterior epidural space pushing the spinal cord anteriorly and extending to the right neural foramina. The arrow points to the tumoral extension to the right neural foramina. The solid arrow points to the anteriorly displaced spinal cord; (b) sagittal post-contrast T1 fat-suppressed image shows brilliantly enhancing mass (large arrow) in the posterior epidural space adjacent to the enhancing hemangioma in posterior aspect of D4 vertebral body (small arrow). The hemangioma also involves the posterior elements (solid arrow)

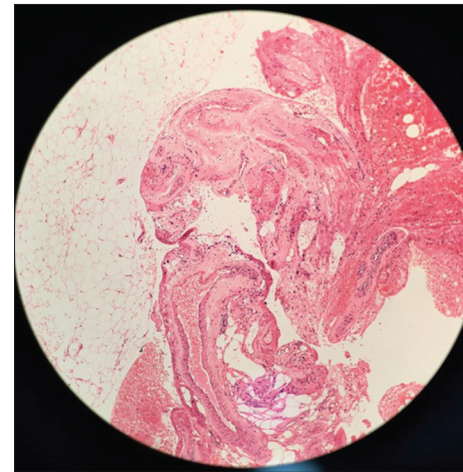


Figure 3: Microscopic hematoxylin and eosin stain shows vascular lesion with huge and medium-sized dilated vessels with thin walls. Surrounding adipose tissue is unremarkable. Consistent with vascular hemangioma

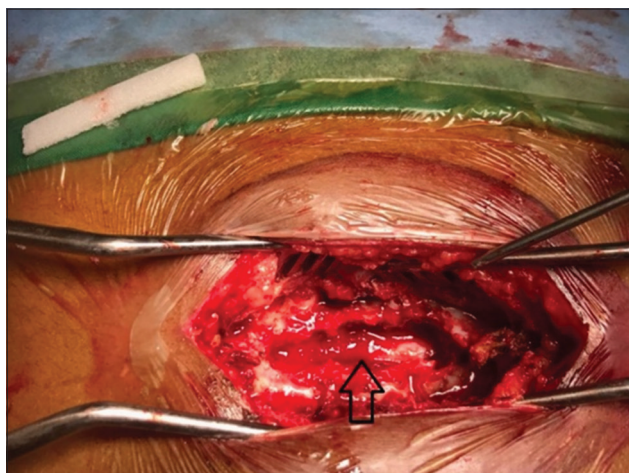


Figure 2: Intraoperative image with arrow pointing at the decompressed spinal cord after laminectomy

DISCUSSION

Vertebral hemangiomas are benign vascular malformations. They can be solitary or multiple. They are usually asymptomatic. The term aggressive hemangiomas are reserved for a small subset (1% of all the hemangiomas), which are symptomatic and are characterized by bone expansion, extraosseous extension, and occasionally fractures [3].

They may present acutely with back pain, myelopathy, or radiculopathy which make radiological investigations necessary. Symptoms may be back pain, acutely and sub-acutely progressive paraplegia, and sensory loss. Neurological symptoms can manifest because of compression fracture of vertebra, sudden hemorrhage in the extradural space, hypertrophy of the posterior vertebral cortex or enlargement of posterior elements due to



Figure 4: (a) Axial T2 post-operative image shows residual soft tissue in the posterior epidural space involving the right neural foramina; (b) sagittal T2 post-operative image shows small residual soft tissue in the epidural space

the extension of the hemangioma, and spinal cord compression due to the extradural expansion of the hemangioma. Aggressive hemangioma most commonly occurs in the D3-D9 vertebral level [4]. When they occur in the thoracic vertebrae, they are more likely to be symptomatic because of the narrow vertebral canal dimensions.

Pregnancy-related symptoms have been known with the potential for rapid symptom progression to myelopathy or cauda equina syndrome due to epidural mass effect. They usually occur in the third trimester [5]. Most of them become asymptomatic post-delivery. The physiological vascular, hemodynamic, and hormonal changes during pregnancy cause a pre-existing hemangioma to enlarge and cause symptoms [6]. By the third trimester, the gravid uterus compresses the vena cava causing an obstruction. Hence, as the uterus increases in size, this venous obstruction and increased intra-abdominal pressure causes increased flow in the vertebral venous plexus, causing expansion and growth of the previously existing hemangioma [6]. This

is the most important cause of the clinical manifestation of pregnancy-induced symptoms. The hormonal changes cause growth-promoting effect mainly through structural changes in the vessel wall in the hemangioma. Maternal progesterone causes an increase in venous distensibility. The endothelial growth-promoting effect of estrogen causes an increase in the size of a pre-existing hemangioma. Relaxin possibly also has an effect [6].

Till now, 26 cases of aggressive hemangioma in pregnancy have been reported [7], but very few cases (two cases) presented the occurrence of the entity in the postpartum period [7]. Ours is a rare case of aggressive vertebral hemangioma presenting in the postpartum period. Jain *et al.* reported a case of progressive spastic paraparesis in the postpartum period due to extradural extension of vertebral hemangioma [5]. Another case of postpartum paraparesis was reported by Mohanty *et al.* where aggressive vertebral body hemangioma was the culprit [8].

Vertebral hemangiomas are characterized by the thickening of vertically striated trabeculae. This appearance has been described on computed tomography as “polka dot,” “corduroy cloth” sign with an overall decrease in the marrow density due to the presence of fatty marrow [9]. Intralesional fat in hemangiomas causes increased signal in T1-weighted MRI images. Aggressive hemangiomas contain less fat and more vascular stroma and thereby have decreased signal on T1 [10]. While the extraosseous component may be hypointense relative to the marrow on non-contrast T1-weighted sequence, uniform enhancement and T2 hyperintensity of osseous and extraosseous part are typical [10]. Cortical erosion, extradural soft-tissue expansion of the posterior elements, and invasion in the spinal canal are radiologic signs of aggressiveness [11]. The diagnosis is easily made but sometimes the differential of malignant lesions such as myeloma and lymphoma metastasis can come. Malignant bone changes such as erosions or aggressive periosteal reaction and accompanying typical features like trabecular coarsening help to distinguish aggressive hemangiomas from these differentials.

Surgery is the treatment of choice of rapidly progressive symptoms of compressive myelopathy and radiculopathy [12] and radiotherapy in case of slowly progressive neurologic deficit. Recently, vertebroplasty, embolization, and intralesional ethanol injection are also being used for the management of symptomatic hemangiomas.

CONCLUSION

Aggressive vertebral hemangiomas constitute 1% of vertebral hemangiomas. Symptoms are mainly due to bone expansion and extraosseous extension. They can manifest during pregnancy due to hormonal and hemodynamic changes and also due to raised intra-abdominal pressure. However, very rarely, as in this case, they can be symptomatic in the postpartum period.

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