

# Symptomatic Spinal Hemangioma in Pregnancy

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**H**emangiomas are among the most common benign tumors involving the spine, usually presenting as an incidental finding on magnetic resonance imaging or even on plain radiography [1]. First described by Virchow in 1867, this lesion is estimated to have an overall incidence of 10–12%, as based on several cadavers and spine film reviews [2]. The lesion may involve any portion of the spinal column, but is more prevalent in the thoracic and lumbar spine segments. The mechanism of growth underlying this often slowly developing lesion is not fully understood. Pregnancy, however, is a well-documented risk factor accompanying its symptomatic conversion, often necessitating surgical decompression.

We report the case of a spinal hemangioma during pregnancy, with unusually severe intra- as well as postoperative complications.

## PATIENT DESCRIPTION

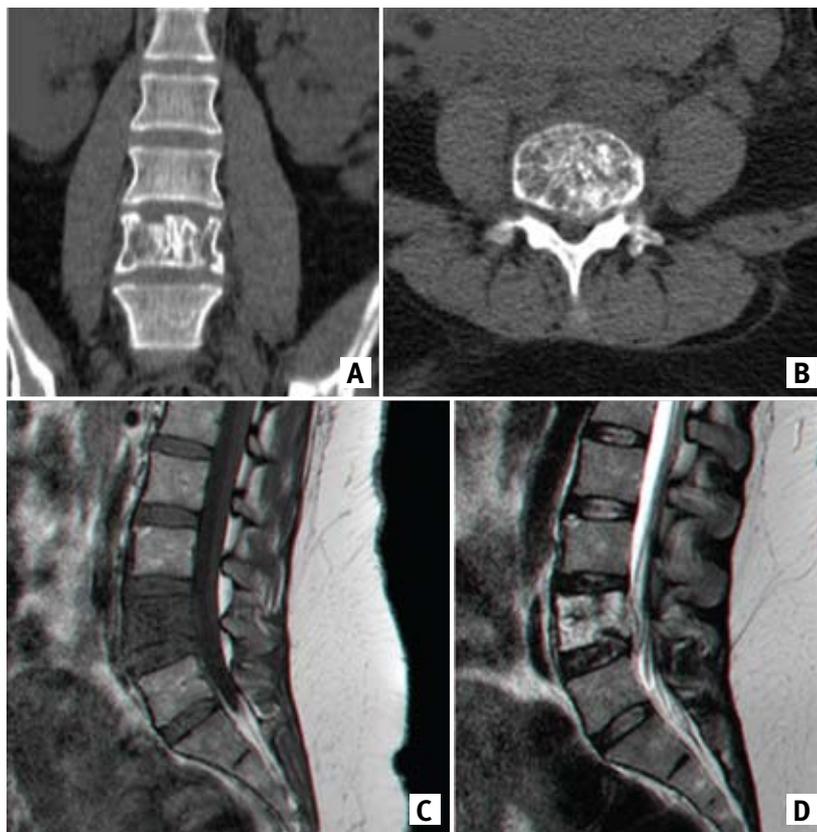
A 35 year old woman presented to the emergency room in the 37th week of gestation with complaints of right abdominal and low back pain. Apart from a history of non-specific low back pain, the patient recalled that 2 weeks previously she had felt a sudden painful snapping sensation in her back that had progressively worsened and began to radiate to her left leg. Diagnosis of premature rupture of membranes led to an

attempt to induce delivery. However, due to failure of both mechanical and pharmacologic means to bring on an active labor over a 48 hour period, together with the patient's overall exhausted state, a cesarean section was performed under general anesthesia.

The operative course was uneventful. Since her complaints persisted postoperatively, she was referred to an orthopedic consultation. Initial physical examination revealed diffuse tenderness in the regions of the lumbar spine and left buttock.

Neurologic examination was marked by a 3/5 weakness in her left tibialis anterior muscle. Lumbar spine X-rays did not demonstrate any pathology. Computed tomography scan revealed a lesion at the fourth lumbar vertebra, marked by lytic and sclerotic areas as well as vertical striations centered in the vertebral body. Soft tissue involvement of the left lateral recess and compression of the thecal sac and the L4 nerve root were also noted [Figure]. A differential diagnosis at this point included a vertebral hemangioma

Anterior-posterior [A] and lateral [B] computed tomography radiographs; T1 [C] and T2 [D] weighted magnetic resonance images of the lumbar spine demonstrating the lesion in the L4 vertebra.



or another primary space-occupying lesion. An additional evaluation, which was carried out to rule out a possible primary malignancy, e.g. multiple myeloma or breast cancer, was negative. Magnetic resonance imaging confirmed the presence of a lesion in the L4 vertebral body, characterized by a mostly low signal intake on T1 and a high signal intake with hypo-intense striated foci on T2-weighted images [Figure].

On admission to the orthopedic department, a standard anticoagulation regimen consisting of low molecular weight heparin, 40 mg daily, was initiated since the persistence of her back pain had resulted in decreased ambulation.

Four days after admission, routine physical examination was marked by rest tachypnea resulting in respiratory decompensation. A clinical suspicion of an acute thromboembolic event was confirmed by computed tomography angiography with a demonstration of an embolus involving the right posterobasal pulmonary segment. Evidence of deep vein thrombosis on ultrasonography led to the installation of an inferior vena cava filter and initiation of full dose anticoagulation therapy.

A week after admission and after a 24 hour cessation of anticoagulation treatment, an attempted percutaneous transpedicular biopsy was performed. During the procedure, bleeding that seemed to originate from vertebral body arteries could only be controlled with hemostats. After failure to attain a biopsy, the patient was discharged and a more thorough tissue-sampling procedure was planned. An outpatient assessment of her coagulation functions did not reveal any known pathologies.

The patient was readmitted several weeks later for an open biopsy of the lesion and decompression of the painful and compressed L4 nerve root. Prior to the procedure, a percutaneous metallic coil embolization was performed by identifying and obstructing the lesion's nourishing arteries. The following day, an open laminectomy and foramino-

tomy at the level of L4 vertebrae enabled a broader retention of tissue sample and decompression of the compressed nerve root. An L4 vertebroplasty concluded the procedure. The postoperative course was uneventful. On discharge, the patient reported a slight improvement in the level of back pain. One year after the procedure, the patient has almost complete resolution of her leg weakness and has satisfactorily returned to her prior level of activity.

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### COMMENT

Hemangiomas are the most common benign vascular bone tumor and usually involve the spinal column and skull. The first known report of a spinal hemangioma in pregnancy is ascribed to Balado in 1927 [3]. In routine practice, the radiographic appearance on CT demonstrating the typical "honeycomb" pattern consisting of vertically oriented striations usually delivers the diagnosis with no need for further evaluation. Exceptions, as portrayed in the current case report, include a non-typical radiographic appearance (e.g. soft tissue involvement, pathologic fracture or compression of neural elements) and significant neurologic impairment.

Hormone regulation and hemodynamic factors are considered leading factors in the lesion's rapid growth during pregnancy. Locally, the enlarging third-trimester uterus increases the pressure in the paravertebral veins by compressing the vena cava. In addition, increased venous distension and vascular growth, thought to be mediated by the systemic influence of progesterone and estrogen respectively, might further increase local venous pressure. These two mechanisms appear to act in synergy on the lesion's close environment, adding to the probability of a compression effect on adjacent neural structures.

Symptomatic conversion leading to neurologic compromise is believed to be due to several pathologies. Direct vascular expansion that originates from the

vertebral elements, mainly the vertebral body, is the most common mechanism. Other mechanisms, such as an enlarging extradural mass (e.g. bleeding) and pathologic fractures, both result in compression of neural elements.

The case presented here has several unique characteristics. Anatomically, most of the previously described symptomatic hemangiomas related to pregnancy were found to involve the thoracic, followed by cervical segments. Reports of lumbar involvement are rare, particularly of pregnancy-related symptomatic lesions.

Clinically, a life-threatening embolic event complicating a diagnosis of an osseous hemangioma is rare. Possible causes for such an event can be classified as being either primary or secondary. Although a rare coincidence of soft tissue angiomas and a consumptive coagulopathy has been described (Kasabach-Merritt syndrome), no direct evidence linking osseous hemangiomas and other clinically significant coagulopathies were found. We hypothesize that two known predisposing factors were involved in the acute embolic event: the continued immobilization imposed by the painful back pain and the general hypercoagulable state of late pregnancy and the puerperium.

Management of acute neurologically disabling lesions in the pregnant woman has been a subject of considerable debate. In addition to surgical decompression, other treatment alternatives include radiotherapy, percutaneous sclerotherapy by ethanol injection and arterial embolization, the latter being supported by a large body of evidence as an important adjunct measure in controlling local bleeding [4].

A management algorithm based on a literature review [5] highlighted two main key aspects in the decision-making process of such scenarios: the week of gestation and the severity of the neurologic impairment. Conservative management, consisting of rest and pain control, was offered to patients approaching the end of their pregnancy

(32nd week of gestation or later) or those with only mild or moderate symptoms, whereas any substantial or progressing neurologic deficit was considered an indication for surgical decompression. However, the modality preferred or its timing was not discussed.

Despite the accumulating information on the management of symptomatic hemangiomas during pregnancy, we found no class I data to support any specific modality of treatment, nor the preferred timing for its application. Recommendations for dealing with the hypercoagulable state are also lacking.

In summary, after reviewing the relevant literature and based on our limited experience, we suggest a multi-

disciplinary approach including interventional radiology for the management of symptomatic hemangiomas affecting the vertebral column, including those related to pregnancy. Preoperative arterial embolization should be considered as an adjunct to any therapeutic modality due to its relative high benefit-risk ratio; and given the increased risk of an acute embolic event due to the hypercoagulable state of pregnancy and the accompanying immobilization, anticoagulant therapy should be started regardless of a planned surgical procedure.

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