Spontaneous Spinal Epidural Hematoma During Pregnancy

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ABSTRACT: Background: Spontaneous spinal epidural hematoma is a rare phenomenon that has no distinct etiology. Spontaneous spinal epidural hematoma (SSEH) during pregnancy is extremely rare. We present what we believe to be the fifth reported case of spontaneous spinal epidural hematoma associated with pregnancy in the English literature. Methods: A 31-year-old female presented with acute onset of paraplegia at 32 weeks of pregnancy. The patient had a T2 sensory level and complete paralysis of all lower extremity motor groups. Magnetic resonance imaging of the thoracic spine showed an acute epidural hematoma posterior to the thoracic spinal cord between the second and fourth thoracic vertebrae. Results: The patient was taken to the operating room were her child was delivered by caesarean section. She then underwent a posterior laminectomy and evacuation of a spinal epidural hematoma. Follow-up selective spinal angiography was negative for any vascular malformation. The patient gradually recovered lower extremity function and was independently ambulating at six month follow-up. Voluntary bowel and bladder function returned within four months but twice daily intermittent catheterization remained necessary for excessive post-void residual urine. Conclusions: Spontaneous spinal epidural hematoma in pregnancy is a rare phenomenon. It is postulated that elevated venous pressure associated with pregnancy may be a contributing factor. In the reported cases of SSEH in pregnancy most patients presented with acute symptoms, thoracic location and profound neurological deficits but, with prompt surgical treatment, generally had good long term recovery.

RÉSUMÉ: Hématome épidural spinal spontané pendant la grossesse. Introduction: L'hématome épidural spinal spontané est un phénomène rare, sans étiologie précise, qui survient très rarement pendant la grossesse. Nous présentons ce que nous croyons être le cinquième cas rapporté d'hématome épidural spinal spontané associé à la grossesse dans la littérature de langue anglaise. Méthodes: Une femme âgée de 31 ans a consulté pour une paraplégie aiguë à 32 semaines de grossesse. La patiente avait un niveau sensitif situé à D2 et une paralysie complète de tous les groupes moteur au niveau des membres inférieurs. L'imagerie par résonance magnétique de la moelle épinière thoracique a révélé la présence d'un hématome épidural aigu situé derrière la moelle épinière thoracique, entre les deuxième et quatrième vertèbres thoraciques. Résultats: La patiente a subi une césarienne, puis on a procédé à une laminectomie postérieure et à l'évacuation de l'hématome épidural. Une angiographie n'a révélé aucune malformation vasculaire. La patiente a recouvré graduellement la fonction de ses membres inférieurs et pouvait marcher sans aide lorsqu'elle a été revue six mois plus tard. Elle a recouvré le contrôle volontaire des fonctions intestinale et vésicale en 4 mois, mais elle devait avoir recours au cathétérisme vésical deux fois par jour à cause d'un résidu vésical important. Conclusions: L'hématome épidural spinal spontané est un phénomène rare pendant la grossesse. Nous postulons qu'une augmentation de la pression veineuse associée à la grossesse pourrait y contribuer. Chez les cas rapportés, la plupart des patientes consultent pour des symptômes aigus et des déficits neurologiques sévères. L'hématome est généralement situé au niveau thoracique et la récupération à long terme est bonne suite à un traitement chirurgical rapide.

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Spontaneous spinal epidural hematoma (SSEH) is a rare medical condition. Spinal epidural hematomas most frequently occur as a complication of trauma or spinal surgery.^{1,2} Causes of SSEH include arteriovenous malformations, anticoagulant therapy, vasculitis, tumours, coagulopathy and prolonged valsalva maneuver.¹⁻⁷ Spontaneous spinal epidural hematoma can occur without predisposing medical conditions. In these cases it is postulated that vertebral venous plexus pressure changes may be responsible.⁸⁻¹² Spontaneous spinal epidural hematoma during pregnancy are exceptionally rare.^{8, 13-15} To our knowledge, this is the fifth reported case of SSEH associated with pregnancy in the English literature.

MATERIALS AND METHODS

Presentation

A 31-year-old female presented to the emergency department with acute onset of paralysis at 32 weeks of pregnancy. Her

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RECEIVED OCTOBER 18, 2004. ACCEPTED IN FINAL FORM FEBRUARY 3, 2005. Reprint requests to: Michael E.B. Kelly, Division of Neurosurgery, Royal University Hospital, 103 Hospital Drive, Saskatoon, SK, Canada S7N 0W8 symptoms began as severe mid-thoracic back pain after urination. Over the next hour, the patient developed a progressive ascending thoracic sensory level accompanied by severe weakness in the lower extremities and loss of bowel and bladder function. The pregnancy had been uncomplicated with no history of hypertension. The patient had a previous uncomplicated pregnancy and delivery in the past. She was otherwise healthy and taking no medications other than a prenatal vitamin supplement. She had no significant family history and did not drink alcohol or smoke.

Examination

Motor examination at initial assessment, three hours after onset of symptoms, showed 0/5 strength in all muscle groups in the lower extremity. Reflexes in the lower extremities were absent and the plantar response was mute. Sensory examination for light touch and pin prick revealed a T2 sensory level. Proprioception was intact in the lower extremities. Her rectal tone and perianal sensation were absent. Upper extremity neurological examination was normal.

Investigations

Laboratory studies showed a platelet count of 205 10³ cells/mm³, partial thromboplastin time was 24 seconds and International Normalized Ratio was 1.0. Urgent magnetic resonance imaging (MRI) of the thoracic spine was performed and demonstrated an epidural mass from T2-T4 which occupied 50% of the spinal canal. Signal intensity on T1 and T2 images was consistent with acute epidural hematoma. (Figures 1 to 3)

Operation

Upon establishing the diagnosis, the patient was taken urgently to the operating room. Less than eight hours from the onset of her symptoms, under general anesthesia, a healthy 32 week child was delivered by caesarian section. The patient was then positioned prone and a T2-T4 laminectomy was performed. An acute epidural hematoma was evacuated with good spinal cord decompression.

Postoperative Investigations

The histopathological analysis revealed only fragments of clotted blood with some fibrovascular tissue but no specific evidence of vascular malformation. Spinal angiography was performed and was normal.

Postoperative course

No motor activity in the lower extremities was present for the first six post operative days. On the seventh post operative day she was able to move her toes and ankles with grade 2/5 strength of plantarflexion and dorsiflexion. By the eleventh day she developed active hip flexion. She was transferred to a rehabilitation facility three weeks postoperatively and had 3/5 hip flexion, 4+/5 ankle plantarflexion and dorsiflexion. At the time of discharge from the rehabilitation ward, two months postoperatively, she had grade 4/5 strength in most lower extremity muscle groups and was ambulating 50 feet with a wheeled



Figure 1: Sagittal T1-weighted MRI illustrating posterior thoracic spinal epidural hematoma (arrow)

walker. At four month follow-up she was voiding independently but self-catheterizing twice daily due to post-void residual urine. By six months, the patient was able to ambulate independently.

DISCUSSION

Spontaneous spinal epidural hematoma associated with pregnancy are rare. 8, 13-15 Only four previous cases have been described in the literature since 1966 and are summarized in Table 1. The etiology in most cases remains unknown and may be multifactorial. However, it has been suggested that increased venous pressure associated with pregnancy may be an underlying factor. This has been emphasized by other authors who have hypothesized that venous bleeding is the major cause of spontaneous epidural hematoma formation. 8-12

Altered hemodynamics and venous pressure within the epidural venous system during pregnancy may predispose patients to SSEH formation.⁸ As the uterus enlarges during the third trimester, these changes are maximal and may explain the increased frequency of SSEH during the final stages of pregnancy. Furthermore, as pregnancy progresses constipation may occur due to alterations in gut motility and pressure from the enlarging uterus. This may require pregnant females to strain

more often to move their bowels. Prolonged valsalva maneuvers have previously been described as a contributing factor in the development of SSEH.⁴

After reviewing the literature, we believe that pregnant women in the third trimester are more likely to develop more severe neurological deficits associated with SSEH formation. The four patients in the literature presented at 28, 35, 37 and 38 weeks gestation. Patients present with relatively increased venous pressure and the tamponade effect that normally slows epidural bleeding would occur only after a greater volume of blood enters the spinal canal. The resultant spinal cord compression would be greater than in non pregnant patients developing a spinal epidural hematoma.

Including our case, four of five patients in the literature presented with acute onset of symptoms.¹³⁻¹⁵ All of these patients had rapid development of significant neurological deficits within several hours of symptom onset. All were found to have cord



Figure 2: Sagittal T2-weighted MRI illustrating posterior thoracic spinal epidural hematoma (arrow)

compression from the mass effect of an epidural hematoma. Steinmetz et al.⁸ described a case with a more subacute onset of symptoms that was not treated with emergent evacuation.⁸

After reviewing a series of SSEH in non pregnant patients, Foo¹⁷ suggested a tourniquet affect from venous bleeding in the

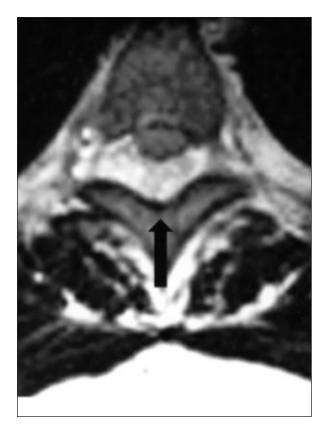


Figure 3: Axial TI-weighted MRI illustrating posterior thoracic spinal epidural hematoma (arrow)

epidural space may be protective and allow for less urgency for surgical evacuation. We believe this may not apply to pregnant patients as vertebral venous pressure is elevated due the effects of pregnancy. This elevation may overcome any tamponade or "tourniquet" effect.

In all reported cases pain has been the initial presenting symptom. The location of pain has been predictive of the level of the hematoma. 8, 13-15 This unique feature should alert those caring for pregnant females as to the potential significance of acute axial pain during pregnancy. These patients should have a thorough neurological examination and be followed closely. If any neurologic abnormality develops, urgent referral to a spine surgeon is required. All patients should have a coagulation profile performed. An MRI should be urgently obtained to establish the diagnosis. Magnetic resonance angiography (MRA) may be performed preoperatively to assess for an arteriovenous malformation. Computerized tomography, myelography or plain x-ray imaging are less likely to be of diagnostic value.

Reviewing the literature and including our case, four of five patients presented with an upper to mid-thoracic epidural hematoma.^{8, 13-15} In two previously reported cases and our case, the hematoma was posterior to the spinal cord.^{13,15} In one of these cases the hematoma was in the cervical spine.¹⁵ Two cases of thoracic hematoma were ventral to the spinal cord.^{8,14} It is currently unclear why there is a tendency towards formation in

Table: Review of published cases of spontaneous spinal epidural hematoma associated with pregnancy in the English literature

Author	Year of publication	Initial Symptom	Weeks Gestation	Onset of symptoms	Location of hematoma	Neurological deficit	Recovery
Bidzinski et al ¹³	1966	Pain	28 weeks	Acute	T2-T5	Complete	Complete
Yonekawa et al ¹⁵	1975	Pain	37 weeks	Acute	C4-C6	Profound	Poor
Carroll et al ¹⁴	1997	Pain	35 weeks	Acute	Т6	Profound	Nearly complete
Steinmetz et al ⁸	2003	Pain	38 weeks	Subacute	T1-T2	Partial	Nearly complete
Kelly et al (Current study)	2005	Pain	32 weeks	Acute	T2-T4	Partial	Complete

the thoracic spine. In a review of 330 cases of SSEH in nonpregnant patients, Groen¹⁸ has suggested no correlation could be observed between the spinal level and postoperative outcome.

It has been described that rapid development of symptoms and severe neurological deficits at presentation are poor prognostic factors for recovery after spinal epidural hematoma formation. ^{10, 17, 18} This is particularly true for lesions affecting the thoracic spinal cord due to smaller canal diameter and greater sensitivity to compressive lesions. ^{18,19} Ultimate outcome after spinal cord decompression has been shown to be related to the preoperative neurological deficit and the operative interval between onset of symptoms and decompression. ¹⁶ The extent of the hematoma (number of spinal segments involved) does not appear to correlate with preoperative neurological deficit or with postoperative outcome. ¹⁸ The patient described by Yonekawa et al ¹⁵ had a worse outcome. This patient was noted to have a severe post operative deficit, a cervical hematoma at C4 to C6 and significant spinal cord edema at surgery.

Because of a paucity of literature on the subject, firm management guidelines cannot be established. The majority of cases in the literature presented with severe, acute neurological deficits. Represented with severe acute neurological deficits. In all cases of thoracic spinal epidural hematoma associated with pregnancy a good clinical outcome was seen with surgical treatment. In one case from 1975, the outcome after cervical spinal epidural hematoma was poor. In patients with profound neurological deficits, aggressive surgical treatment is warranted as significant neurological recovery can be achieved with prompt decompression.

In cases of acute spinal epidural hematoma associated with pregnancy, we believe that once the diagnosis is established the most appropriate treatment is prompt surgical evacuation of the hematoma. Surgical decision making should involve a multi-disciplinary team including members from obstetrics, anesthesia and spine surgery. Fortunately most cases of SSEH during pregnancy occur in the third trimester. This allows for delivery of the baby by caesarian section prior to embarking on spine

surgery. In less advanced pregnancies, when gestational age would preclude delivery of the child, discussions must be undertaken between the patient, obstetrician and spine surgeon as to potential risks to the fetus. The surgical approach must be tailored to the location of the hematoma with appropriately positioned midline skin incision, laminectomy and hematoma evacuation. Steinmetz et al⁸ describe the option of lateral positioning and anterolateral or posterolateral approaches in patients in the first and second trimester without delivery of the fetus.

Postoperative management should be expectant with appropriate post partum care in addition to management of a neurologically impaired patient. Issues such as deep venous thrombosis prophylaxis, bowel and bladder management and mobilization should be addressed in combination with obstetrical staff and include early involvement from rehabilitation medicine specialists. All reported cases of spinal epidural hematoma associated with pregnancy have been spontaneous and the utility of further imaging with spinal angiography or MRA to rule out arteriovenous malformation remains unclear.

Spontaneous spinal epidural hematoma associated with pregnancy is a rare occurrence. The etiology of this condition is unknown although venous hypertension associated with pregnancy is a possible etiology. Most patients present with pain localized to the level of the hematoma and acute onset of severe neurological deficits. Prompt diagnosis and surgical evacuation allows a good function outcome in most patients including those with severe preoperative neurological deficits.

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