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Spontaneous thoracic epidural hematoma after normal vaginal delivery: a case report and literature review

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Summary

Spontaneous spinal epidural hematoma (SSEH) is an extremely rare complication in the postpartum period. SSEH in thoracic spine after natural delivery has never been reported. A 25-year-old women presented with acute onset of paraplegia eight hours after normal vaginal delivery. MRI revealed a right lateral mass in the thoracic spinal canal extending from T1 to T2. Prompt decompressive laminectomy was performed within 12 hours after the event. Follow-up at one month post-operation showed a favorable neurological outcome of the patient. In addition, literatures of similar SSEH cases during pregnancy and postpartum period were reviewed, aiming to highlight the importance of early recognition and expedient intervention of SSEH.

Key words: Spontaneous spinal epidural hematoma; Thoracic spine; Paraplegia; Pregnancy; Postpartum.

Introduction

Spontaneous spinal epidural hematoma (SSEH) was firstly described by Jackson in 1869 and first surgical treatment was reported by Bain in 1897 [1, 2]. SSEH is a uncommon idiopathic condition that results in sudden onset of neurological deficit. It is an extremely rare complication in pregnancy and postpartum period. Only one SSEH case in cervical spine after natural delivery was reported before [3]. However, SSEH in thoracic spine after normal vaginal delivery has never been reported. The early recognition and adequate surgical treatment are of great importance to prevent permanent neurological consequences and to accelerate postoperative recovery. The authors herein present a unique case of SSEH in a woman who developed paraplegia eight hours after natural delivery. The case was unique in: 1) quick onset with hematoma in thoracic spine, 2) right lateral hematoma, and 3) onset after natural delivery. Retrospective review of other similar cases was also done to discuss the etiology, presentation, and management of SSEH.

Case Report

The patient, a previously healthy 25-year-old woman, presented to the authors' department with progressive paraplegia after giving birth to her first child by normal vaginal delivery. She was initially well observed in the obstetric ward of local hospital until eight hours of the postpartum period when she complained of a sudden onset of anesthesia and weakness of lower limb. She was seen by a physician in the local hospital who prescribed mannitol but without remarkable improvement. Within one hour, severe weakness in both legs developed along with urinary incontinence and sensory disturbance from the nipple line downward. Due to progressively worsening symptoms of motor and sensory deficits, she was immediately transferred to the authors' unit for diagnosis and treatment. She had no history of vomiting, fever, headache, photophobia, or seizure. She denied medical history of recent infection, trauma, homeostasis diseases (inherited or acquired), preexisting neurological deficit, taking anticoagulants, or receiving invasive manipulations such as epidural analgesia. On admission, she had a Glasgow Coma Scale (GCS) of 15/15. Pupils were normal and sensitive to light reflex. She had a body temperature of 37.2°C, blood pressure of 113/79 mm Hg, and heart rate of 78 beats per minute. Physical examination revealed paraplegia from the level of T2, with lower limb power at Grade 0/5 both proximally and distally. Her sensation was diminished in both lower extremities, but proprioception was intact. Babinsky reflex was positive on both sides. Physical examination on cranial nerves and upper limbs was normal.

The patient's coagulation profile including blood cell count, platelet count, and prothrombin time was unremarkable within normal limits. Plain radiographs on cervical and thoracic spine revealed no significant abnormalities. MRI on the same segment revealed a right lateral, moderate-sized epidural mass extending from T1-T2 with slight signal changes in spinal cord (Figure 1). The signal characteristics of the mass indicated suspicious epidural hematoma. To exclude the possibility of hemorrhage resulted from arteriovenous malformation (AVM), an urgent spinal cord angiography was therefore performed but showed negative results.

Due to the severity of compressive spinal cord, within 12 hours of her admission, an emergency decompressive laminectomy was performed with the patient in a prone position under general anesthesia. Intraoperatively, a partially organized hematoma was identified on the right side of T2 foramen intervertebral compressing

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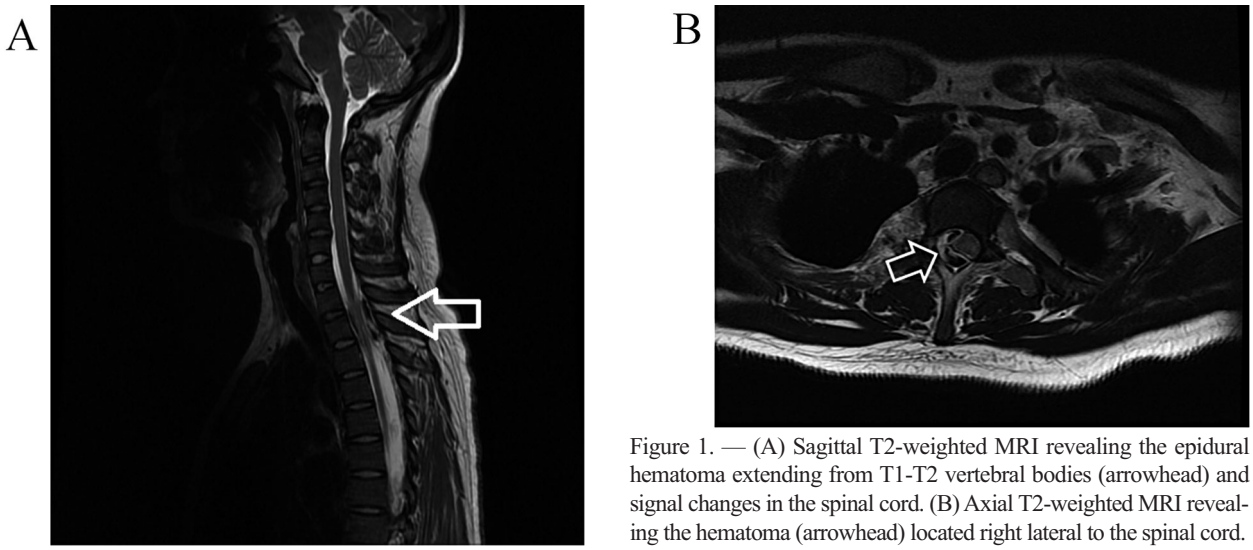

Figure 1. — (A) Sagittal T2-weighted MRI revealing the epidural hematoma extending from T1-T2 vertebral bodies (arrowhead) and signal changes in the spinal cord. (B) Axial T2-weighted MRI revealing the hematoma (arrowhead) located right lateral to the spinal cord.

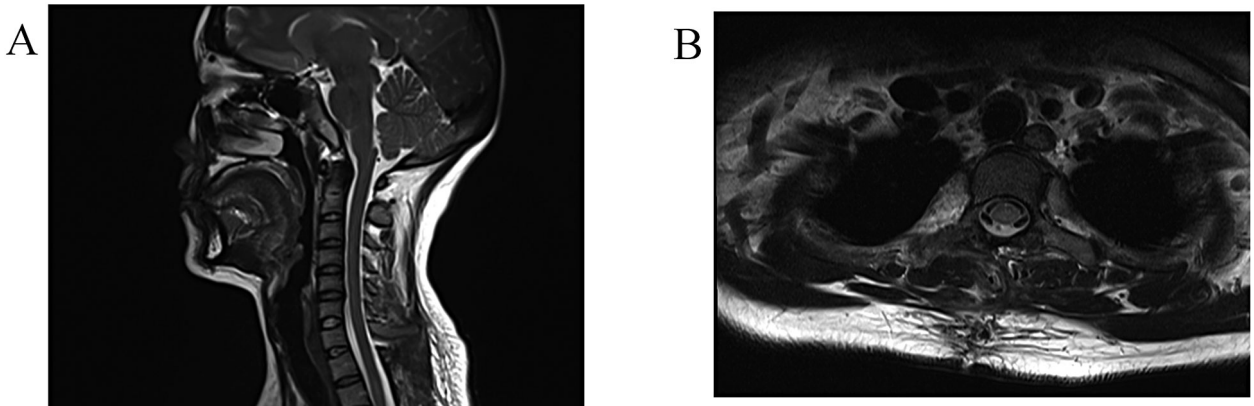

Figure 2. — (A) Sagittal and (B) axial T2-weighted MRI revealing no evidence of any residual hematoma or spinal cord compression postoperation. Pathological enhancement is not detected on T1-weighted contrast MRI image (B).

the spinal cord at T1–T2. There was no evidence of a vascular malformation or bleeding source within the spinal canal. After careful evacuation of the hematoma, spinal cord was freed and a drain was inserted before wound closure.

Postoperatively, power of the patient's both lower limbs witnessed a delightful recovery at Grade 4/5, and sensation was regained rapidly. A follow-up MRI examination one month after surgery revealed complete evacuation of the original epidural hematoma and normal signal of freed spinal cord (Figure 2).

Discussion

SSEH rarely occurs in pregnancy and postpartum period. The present authors searched the English literature to review the reported cases. The summary is organized in Table 1. Only two SSEH cases in postpartum period have been

Table 1. — *Summary of reported SSEH cases in pregnancy and postpartum period.*

Author and year	Onset time	Location of hematoma	Interval from onset to decompression	Outcome and measurement time from decompression
Present study	8 hours after virginal delivery	T1-T2	12 hours	Full recovery, 1 month
Bose S, 2007 [3]	2 weeks after virginal delivery	C4-C7	Surgery not taken	Quadriplegia, 45 days from admission
Puah KL, 2012 [4]	3 weeks after cesarean section	C6-T2	24 hours	Neurological recovery, ladder dysfunction, 1 year
Iwatsuki K, 2015 [27]	37 weeks of pregnancy	C4-T1	None	Spontaneously resolved after cesarean section
Wang Z, 2013 [28]	26 weeks of pregnancy	T6-T10	10 days	Paraplegia, 6 months
Jo YY, 2012 [29]	36 weeks of pregnancy	T1-T5	< 15 hours	Full recovery, 37 days
Wang P, 2011 [7]	40 weeks of pregnancy	C5-C7	< 18 hours	Ambulatory, 6 months
Tada S, 2011 [30]	Case 1: 31 weeks of pregnancy Case 2: 39 weeks of pregnancy Case 3: 36 weeks of pregnancy	Case 1: C4-T4 Case 2: C4-T2 Case 3: T5-T8	Case 1: 30 hours Case 2: 12 hours Case 3: 8 hours	Case 1: Full recovery, 3 months Case 2: Good recovery, 1 month Case 3: Full recovery, 10 days
Badar F, 2011 [19]	37 weeks of pregnancy	D2-D8	3 days	Good recovery, 2 weeks
Matsubara S, 2011 [31]	16 weeks of pregnancy	C3-C7	9 hours	Quadriplegia, 13 weeks
Forsnes E, 2009 [32]	27 weeks of pregnancy	T12-L2	16 hours	Ambulatory, 17 weeks
Singh DP, 2009 [33]	31 weeks of pregnancy	C3-C7	30 hours	Full recovery, 15 days
Jea A, 2005 [34]	20 weeks of pregnancy	T1-T2	8 hours	Complete neurological recovery, 1 year
Case AS, 2005 [35]	37 weeks of pregnancy	T6-T9	10 hours	Full recovery, 1 year
Doblar DD, 2005 [6]	37 weeks of pregnancy	T6-T9	11.5 hours	Ambulatory, 8 months
Kelly MEB, 2005 [36]	32 weeks of pregnancy	T2-T4	< 8 hours	Ambulatory, 6 months
Szkup P, 2004 [11]	32 weeks of pregnancy	T1-T4	< 7 hours	Ambulatory, 10 months
Cywinski JB, 2004 [5]	38 weeks of pregnancy	T1-T2	> 36 hours	Ambulatory, 8 weeks
Masski G, 2004 [37]	41 weeks of pregnancy	C7-T2	12 hours	Paraplegia, 2 months
Steinmetz MP, 2003 [10]	38 weeks of pregnancy	T1-T2	> 36 hours	Ambulatory, 3 days
Carrol SG, 1997 [12]	35 weeks of pregnancy	T6-T7	< 24 hours	Ambulatory, not mentioned
Yonekawa Y, 1975 [38]	35 weeks of pregnancy	C4-C6	17 hrs	Paraplegia, 11 months
Bidzinski J, 1966 [39]	25 weeks of pregnancy	T2-T5	31 hours	Ambulatory, 8 weeks

SSEH: spontaneous spinal epidural hematoma.

reported previously in English literature [3, 4]. Bose *et al.* reported the first SSEH case after natural delivery in 2007 and the hematoma occurred in cervical spines [3]. In this paper the authors reported the first thoracic epidural hematoma case after natural delivery. Puah *et al.* reported one SSEH case in cervical spines after cesarean section [4]. Twenty literatures were summarized and reported 22 SSEH cases during pregnancy in Table 1. Due to the language issue, the authors did not include details of other possible cases. The estimated sum of SSEH cases during pregnancy might be approximately 30 based on the literatures the present authors have read. Some previous references have provided information on those cases reported in other languages in addition to English [5-7]. In Puah *et al.*'s report, whether epidural analgesia was performed before the cesarean section is not mentioned [4]. Iatrogenic procedure like epidural analgesia was considered to be traumatic and might confuse the diagnosis of SSEH [8]. Owing to the absence of accurate definition by far, it is questionable whether all the cases reported are to qualify the final diagnosis of SSEH.

The exact etiology for SSEH is universally unclear. Recognized elements that could predispose to SSEH include anticoagulant therapy, vascular malformations, hemophilia, and vasculitis [9, 10]. In the present case, the possible risk factors as mentioned above were ruled out through examination. Therefore the present case might have been caused by perinatal factors. Based on the widely accepted venous origin hypothesis, epidural veins in the spine are thin-walled and valveless, making the spine more vulnerable and subsequently leading to rupture owing to increased thoracic, abdominal, and pelvic pressure during pregnancy [11-13]. It is notable that uterine contraction and redistribution of interstitial fluid after pregnancy can cause a remarkable increase of body circulation [14]. The combination of hormonal changes on vascular walls and rapid increase of blood volume may lead to further hemorrhage. A relative hypercoagulable state of puerperium may then account for difficulty in absorption of hematoma. However, the rapid development of hematoma in some cases also suggested an arterial origin. According to Beatty's hypothesis, hemorrhage in cervical and upper dorsal cord was arterial in origin because bleeding from a low-pressure venous system could not expand very quickly and

cause compression [15]. Gopalkrishnan *et al.* supported Beatty's theory by summing up that high mobility of C6-C7 may further account for this common location of SSEH [16]. Based on the present literature review, ten out of 25 cases had hematoma involving C6-C7 segment, and the other 15 cases had hematoma expanding from thoracic spine and below. The results indicate that the exact etiology of SSEH still needs further investigation.

Local spinal cord compression but not vascular obstruction is thought to be the main factor causing neurologic problems, so expedient surgical intervention is crucial for favorable prognosis of SSEH. Based on multiple reviews, good recovery was usually related with short interval (within 36-48 hours) from onset to decompression intervention [17, 18]. Occasionally a number of SSEH cases reported satisfactory outcomes on conservative management, which might be explained by obviously smaller lesion size and minimal symptoms or signs [13, 19, 20]. Bose *et al.* also recommended to prolong follow-up time before announcing the final outcome because sometimes it might take months to recover [3].

Careful observation in obstetric ward is therefore important to recognize early symptoms of SSEH in puerperium stage. Clinical manifestations for SSEH include a sudden onset accompanied with apparent pain in the chest-back region and acute symptoms, such as limb weakness, paraplegia, and urinary retention. Owing to varied locations and ranges of epidural hematoma, motor and sensory deficits are closely correlated with the level and degree of the spinal cord compression. This lesion can also be atypical or progressively increase in severity by presenting conditions of Brown-Sequard syndrome, quadriplegia, or respiratory paralysis [3, 21]. SSEH lower than cervical spine tends to have a subacute or chronic course [16]. Bose *et al*. and Puah *et al*. each reported a case with slow progression in the postpartum period [3, 4]. Their patients developed multiple neurological deficit within two and three weeks, respectively. The present illustrative case is unique in that it suggests SSEH in thoracic spine may also develop quickly (eight hours after virginal delivery).

A prompt MRI is particularly useful in the early stages by presenting convincing evidence for the diagnosis of SSEH. The signal intensity of the SSEH is usually isointense on T1-weighted images and hyperintense on T2-weighted images. Small heterogeneous patchy hypointensity on T2-weighted images within the lesion suggests an acute spinal epidural hematoma [13]. By review of literatures, Gopalkrishnan *et al.* concluded that SSEH was mostly located dorsal to spinal cord, which demonstrates that the right lateral hematoma in the present case is even rare [16]. To rule out suspicious SSEH resulting from AVM, a spinal cord angiography, as was performed in the present case, can be a useful approach given the fact that escaped blood flow signal on MRI may confuse the diagnosis between AVM and hematoma. Differential diagnosis for SSEH includes multiple sclerosis (MS) and neuromyelitis optica (NMO), both of which are identified to have an increase in onset and relapse risk in early postpartum period, and are more prevalent in Asia countries like Japan and China [22-25]. The two demyelinating diseases of central nervous system can present with an acute or subacute onset of limb weakness and sensory deficits similar to SSEH, but they can be differentiated via radiological evidences and specific markers in cerebrospinal fluid (CSF) investigations, such as CSF-lgG oligoclonal bands (OB) or aquaporin-4 (AQP4) antibodies [26].

Conclusion

SSEH is rare in postpartum period. The present authors reported the acute onset of thoracic hematoma in a women and good recovery was achieved after timely decompression. The etiology and pathophysiology course remain unclear to our knowledge. Obstetricians and neurosurgeons should be aware of a pregnant woman presenting sudden onset of neck or back pain with signs of a compressive spinal cord, even after normal vaginal delivery. An urgent MRI is useful to identify, differentiate, and locate SSEH. To prevent irreversible neurological consequences and accelerate postoperative recovery, the interval between early diagnosis and urgent surgical treatment should be shortened as much as possible.

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