# Spontaneous Spinal Epidural Hematoma: A Case Report and Review of the Literatures

Chung-Chen Lo<sup>1</sup>, Jun-Yih Chen<sup>2</sup>, Yuk-Keung Lo<sup>1,4</sup>, Ping-Hong Lai<sup>3,4</sup>, Yuh-Te Lin<sup>1,4</sup>

#### Abstract-

- *Purpose:* To emphasize the importance of early recognition and emergent surgery for spontaneous spinal epidural hematoma (SSEH).
- *Case Report:* A 61-year-old female presented with sudden onset of severe neck and back pain after finishing worshiping Buddha followed by quadriparesis, sensory deficits below C4 level and sphincter dysfunction. MR imaging demonstrated acute extensive epidural hematoma of cervico-thoracic spinal segments (C2-T7). Idiopathic SSEH was diagnosed and emergent decompressive laminectomy with hematoma evacuation was performed within 12 hours of symptoms onset. Good functional and neurological outcomes were obtained.
- *Conclusion:* SSEH is a rare but disabling or even fatal entity. Early diagnosis and prompt surgery improve the neurological and functional outcome but still remain a clinical challenge. Relevant physicians should pay attention to the typical symptoms of the rare entity and SSEH should be one of differential diagnoses.
- Key words: spinal epidural hematoma, spinal cord compression, magnetic resonance imaging, neurosurgery

Acta Neurol Taiwan 2012;21:31-34

## INTRODUCTION

Despite Jackson reported the first case of spontaneous spinal epidural hematoma (SSEH) early in 1869 <sup>(1)</sup>, SSEH is still a rare but important cause of spinal cord compression in the emergency department. The incidence of SSEH has been estimated at 0.1 patients per 100,000 individuals and represents less than 1% of spinal space-occupying lesions<sup>(2)</sup>. SSEH usually presents with sudden onset of neck or back pain followed by symptoms and signs of rapidly evolving nerve root and spinal cord compression. The early diagnosis and prompt management correlate with good outcome but still remain a challenge for physicians. Here, we report a case of idiopathic SSEH with most extensive levels of cervico-thoracic spinal segments (C2-T7) and review the relevant literatures.

Correspondence to: Yuh-Te Lin, MD, PhD. Section of Neurology, Kaohsiung Veterans General Hospital, No. 386, Ta-chung 1st Road, Kaohsiung, Taiwan. E-mail: ytlin@isca.vghks.gov.tw

From the <sup>1</sup>Section of Neurology, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan; <sup>2</sup>Section of Neurosurgery, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan; <sup>3</sup> Department of Radiology, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan; <sup>4</sup>School of Medicine, National Yang-Ming University, Taipei, Taiwan.

# **CASE REPORT**

A 61-year-old female was brought to the emergency department (ED) with the sudden onset of severe sharp neck and upper back pain, occipital headache, general weakness and transiently disturbed consciousness when she finished worshiping Buddha at home. She fell down to the ground and then above symptoms developed rapidly within 2 hours. She couldn't stand up without assisstance. There was no past history of head and spinal trauma, smoking, drinking nor family history of vascular problems. She had type 2 diabetes mellitus and hypertension under regular medications but she didn't take any antiplatelet or anticoagulant agents. On examination, the patient was alert, orientated and afebrile with a blood pressure of 140/86 mmHg. She had an unremarkable head, neck, chest, cardiovascular and abdomen examination. Neurological examination revealed the muscle



Figure 1. Pre-operative cervical and thoracic MRI of acute SSEH. Sagittal T1-weighted images (Fig. 1a and 1b) show posterior epidural isointense lesion (arrows) and T2-weighted images (Fig. 1c and 1d) show mild hyperintensity over posterior epidural space from C2 to T7 (arrows) with cord compression. power exhibited grade 2-3/5 in left upper limb and 4/5 in other three limbs. Impaired pink-prick sensation below C4 level was noted. Her reflexes were normal. Acute urinary retention developed later and Foley catheter was indwelled at ED. Laboratory investigation including complete blood count, chemistry panel, and coagulation profile were all within normal limits. MR imaging demonstrated acute extensive posterior spinal epidural hematoma from C2 through T7 causing compression of the spinal cord. The acute hematoma was relative isointense on T1-weighted images and mild hyperintense on T2-weighted images (Fig. 1). Emergent decompressive laminectomy from C3 to T3 was performed to remove large epidural hematoma within 12 hours after the onset of symptom. There are neither abnormal vessels nor hemorrhagic tumors seen during operation to explain the bleeding. After operation, muscle strength recovered well with 4/5 over left upper limb and 5/5 in other three limbs. Followed-up MRI of C-spine revealed adequate cord decompression.

### DISCUSSION

Althought spontaneous spinal epidural hematoma (SSEH) is an accumulation of blood in the vertebral epidural space in the absence of trauma or iatrogenic procedure like lumbar puncture, there is still no agreedupon definition. Some authors include hematomas secondary to coagulopathy, vascular malformations and hemorrhagic tumors. The other authors claim, however, that hematoma can be labeled spontaneous only when it is of idiopathic origin<sup>(3)</sup>. Statistically, idiopathic cases accounts for approximate 40% to 61% in the previous studies <sup>(4,5)</sup>. The location and onset age of SSEH have a bimodal distribution with the location peaks at C6 and T12 and onset age peaks at 15-20 and 65-70 respectively <sup>(6,7)</sup>. The gender ratio (male-female) is 1.4:1<sup>(8)</sup>. There are some precipitating factors associated with SSEH, such as anticoagulant therapy for prosthetic cardiac valve<sup>(9)</sup>, thrombolysis therapy for acute myocardiac infarction<sup>(10)</sup>, uncontrolled hypertension<sup>(11)</sup>, end-stage renal disease receiving hemodialysis<sup>(12)</sup>, long-term antiplatelet usage <sup>(13)</sup> and congenital diseases with factor XI deficiency or

hemophilia B<sup>(14,15)</sup>.

The most common initial symptom of SSEH is sudden onset of severe neck and /or back pain often radiating to corresponding dermatome. Motor and/or sensory deficits caused by compression of nerve roots and spinal cord follow and progress within several hours depending on the levels of lesions. In some cases and the present patient sphincter disorders have developed <sup>(16)</sup>. Despite the characteristic syndrome of SSEH, early and accurate diagnosis still remains a challenge for relevant physicians. After the introduction and general usage of MR imaging, the diagnostic capabilities, including early and accurate diagnosis, the location and size of hematoma and edema and compression severity of spinal cord have been improved than before. The early MR images of acute SEH reveal isointense or hypointense on T1weighted images and hyperintense on T2-weighted images. In addition to specific signal changes, contrast enchancement pattern and morphological findings on MR images can differentiate acute SEH from spinal epidural neoplastic mass or abscess<sup>(17)</sup>.

The pathogenesis of SSEH is not clear as to the source of hematoma with literature in support of both venous and arterial origins. Most authors accept the venous etiology hypothesis due to lacks of venous valves in epidural venous plexus. Sudden increasing pressure from the thoracic or abdominal cavity would result in vessel rupture and hemorrhage. Recent cases series showed 54% of SSEH patients reported a straining-associated event during the initial attack <sup>(18)</sup>. Some authors have supposed the spinal epidural artery as a rupture vessel because the pressure in venous plexus is lower than that in epidural space and rapidly deteriorating neurological deficit clinically <sup>(3)</sup>. Further studies are needed to clarify the precise pathogenesis of SSEH.

SSEH is generally surgical emergency with rapid decompressive laminectomy and hematoma evacuation being the most effective treatment <sup>(19,20)</sup>. Conservative treatment is still an important option of treatment in some selective patients with mild and rapidly spontaneous recovery symtoms <sup>(21)</sup> or high sugical risk patients with bleeding tendency associated with severe systemic disease, advanced cardiovascular disease or advanced

and irreversible spinal cord injury. Recent report showed the results of conservative treatment were poor in cases with cervical lesions (22). Approximately, post-operative mortality rate of SSEH is around 3% to 6% (18,23). The prognosis of SSEH correlates with the size and level of hematoma, severity of pre-operative neurological deficits and time interval between symptom onset and surgery. Recent study reveals the common levels of hematoma range between 2 and 10 spinal segments and long hematoma predicts worse outcome<sup>(5)</sup>. Liao et al reports the larger size of SSEH induced by impaired hemostasis shows poor post-operatively functional recovery <sup>(20)</sup>. In the review of literatures, we report the present idiopathic SSEH case with the most extensive levels (13 segments, C2-T7) and without coagulopathy. Previous studies show the surgical outcome of SSEH is inversely related to the time interval between symptoms onset and surgery (5,18,20,23-25). The likelihhood of recovery improved significatly when oprations were performed within 36 and 48 hrs in SSEH pateints with complete and incomplete deficits respectively (18). In Shin's series, the recovery scale of the Japanese Orthopedic Association (JOA) were 83%, 63.6% and 46.7% for the SSEH patients who received surgery less than 12 hrs symptoms onset, between 12 and 24 hrs and after 24 hrs respectively<sup>(24)</sup>. Lawton et al argued that the time interval should be brought forward to 12 hours for better neurological outcomes (23). Liao et al series showed there was a trend toward complete spinal functional loss due to SSEH lasting more than 12 hours predicting permanent disability. Therefore, it is assumed that early surgery within 12 hrs may give SSEH patients with complete deficits the best opportunity for functional recovery<sup>(20)</sup>.

SSEH is a rare but disabling or even fatal condition. Early diagnosis and prompt management improve the prognosis and outcome but still remain a clinical challenge. Relevant physicians should understand the typical symptoms and consider SSEH as one of differential diagnoses.

## REFERENCES

1. Jackson R. A case of spinal apoplexy. Lancet 1869;2:5-6.

- 2. Holtas S, Heiling M, Lonntoft M. Spontaneous spinal epidural hematoma: findings at MR imaging and clinical correlation. Radiology 1996;199:409-413.
- Betty R, Winston K. Spontaneous cervical epidural hematoma: a consideration of etiology. J Neurosurg 1984; 61:143-148.
- Penar PL, Fischer DK, Goodrich I, Bloomgarden GM, Robinson F. Spontaneous spinal epidural hematoma. Int Surg 1987;72:218-221.
- Liu Z, Jiao Q, Xu J, Wang X, Li S, You C. Spontaneous spinal epidural hematoma: analysis of 23 cases. Surg Neurol 2008;69:253-260.
- Kreppel D, Antoniadis G, Seeling W. Spinal hematoma: a literature survey with meta-analysis of 613 patients. Neurosurg Rev 2003;26:1-49.
- Groen RJM. Non-operative treatment of spontaneous spinal epidural hematomas: a review of the literature and a comparison with operative cases. Acta Neurochir (Wien) 2004; 146:103-110.
- Lonjon MM, Paquis P, Chanalet S, Grellier P. Nontraumatic spinal epidural hematoma: report of four cases and review of the literature. Neurosurgery 1997;41:483-487.
- Vaya A, Resureccion M, Ricart JM, Ortuno C, Ripoll F, Mira Y, Aznar J. Spontaneous cervical epidural hematoma associated with oral anticoagulant therapy. Clin Appl Throm Hemost 2001;7:166-168.
- Van Schaeybroeck P, Van Calenberg F, Van De Werf F, Demaerel P, Goffin J, Plets C. Spontaneous spinal epidural hematoma associated with thrombolysis and anticoagulation therapy: report of three cases. Clin Neurol Neurosurg 1998;100:283-287.
- Spengos K, Tsivgoulis G, Zakopoulos N. Could high blood pressure be the cause of acute spontaneous spinal epidural hematoma? Eur J Emerg Med 2007;14:59.
- Deger SM, Emmez H, Bahadirli K, Kale A, Ebinc FA, Turkoglu M, Arinsoy T, Sindel S. A spontaneous spinal epidural hematoma in a hemodialysis patient: a rare entity. Intern Med 2009;48:2115-2118.
- Weber J, Hoch A, Kilisek L, Spring A. Spontaneous intraspinal epidural hematoma secondary to use of platelet aggregation inhibitors. Dtsch Med Wochenschr 2001;126:

876-878.

- Mustafa MH, Bernstain RA. Spontaneous spinal epidural hematoma, Brown-sequard syndrome, and factor XI deficiency. Ann Intern Med 1987;106:477-478.
- Bisson EF, Dumont T, Tranmer B. Spontaneous Spinal Epidural Hematoma in a Child with Hemophilia B. Can J Neurol Sci 2007;34:488-490.
- Bose S, Ali Z, Rath GP, Prabhakar H.Spontaneous spinal epidural hematoma: a rare cause of quadriplegia in the post-partum period. Br J Anaesth 2007;99:855-857.
- Chang FC, Lirng JF, Chen SS, Luo CB, Guo WY, Micheal MH Teng, Chang CY. Contrast enhancement patterns of acute spinal epidural hematomas: A report of two cases. AJNR 2003;24:366-369.
- Liao CC, Lee ST, Hsu WC, Chen LR, Lui TN, Lee SC. Experience in the surgical management of spontaneous spinal epidural hematoma. J Neurosurg 2004;100:38-45.
- Matsumura A, Namikawa T, Hashimoto R, Okamoto T, Yanagida I, Hoshi M, Noguchi K, Takami M. Clinical management for spontaneous spinal epidural hematoma: diagnosis and treatment. Spine J 2008;8:534-537.
- 20. Liao CC, Hsieh PC, Lin TK, Lo YL, Lee SC. Surgical treatment of spontaneous spinal epidural hematoma: a 5year experience. J Neurosurg Spine 2009;11:480-486.
- Hentschel SJ, Woolfenden AR, Fairholm DJ. Resolution of spontaneous spinal epidural hematoma without surgery. Spine 2001;26:E525-E527.
- 22. Chan DT, Boet R, Poon WS, Yap F, Chan YL. Spinal shock in spontaneous cervical spinal epidural hematoma. Acta Neurochir (Wien) 2004;146:1161-1162.
- 23. Lawton MT, Porter RW, Heiserman JE, Jacobowitz R, Sonntag VK, Dickman CA. Surgical management of spinal epidural hematoma: relationship between surgical timing and neurological outcome. J Neurosurg 1995;83:1-7.
- Shin JJ, Kuh SU, Cho YE. Surgical management of spontaneous spinal epidural hematoma. Eur Spine J 2006;15:998-1004.
- Groen RJ, van Alphen HA. Operative treatment of spontaneous spinal epidural hematomas: a study of the factors determining postoperative outcome. Neurosurgery 1997;39: 494-508.