Case Report

Fatally Idiopathic Spontaneous Hemoperitoneum in a Patient with Cervical Spine Injury: A Case Report and Review of the Literature

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Objectives: *Idiopathic spontaneous hemoperitoneum has never been reported in patients with associated orthopedic injury. The present report aimed to demonstrate a case of this life-threatening condition concomitant with cervical spine injury.*
Case Report: *A 50-year-old male was an illegal immigrant and transferred to the emergency department with the conditions of the bilateral facet subluxation of C5-6 spine, including incomplete cord lesion (American-Spinal-Injury-Association: grade-C) and without associated injury, caused by falling from a 3-meters-scaffold. He underwent anterior cervical discectomy, fusion with iliac bone graft, and plating of C5-6 level at 20-hours after injury. His overall conditions recovered gradually with the same neurological deficit as pre-operative status. On postoperative day 15, he developed acute hemodynamic instability and cardiac arrest in an hour without preceding signs. Autopsy showed free intra-peritoneal blood 1,000 ml and fresh blood in gastric lumen-small bowels 500 ml without definitive source of bleeding.*
Discussion: *The patient in this present report had no complaint of preceding abdominal symptoms in both pre-and-postoperative periods. His visceral sensibility might be lost due to incomplete cord lesion. At this point, he might have an incompetency of detection of intra-abdominal abnormality when it occurred. Hence, this patient had no classic symptoms of spontaneous hemoperitoneum. This made the diagnosis of this condition quite difficult in the presented patient. This situation was very uncommon and essentially unique.*
Conclusion: *The authors recommend "high index of suspicion" of this diagnosis as a cause of hemodynamic instability in patients with neurological deficit, due to spine injury or any cause, which might prevent them to recognize antecedent abdominal symptoms.*

Keywords: *Spontaneous hemoperitoneum, Idiopathic, Fatal, Cervical spine, Injury*

J Med Assoc Thai 2011; 94 (7): 882-7
Full text. e-Journal: *http://www.mat.or.th/journal*

Spontaneous hemoperitoneum is an infrequent condition. This condition might be derived from various aetiology such as pregnancy-related, gynecologic, pancreatic, biliary, splenic, liver, gastric, colonic, coagulopathy, and vascular causes[1-9]. Little was known about an idiopathic spontaneous hemoperitoneum. It has been rarely reported in previous literatures[10], especially in patients with orthopedic conditions. Although it is a rare individual, it may be life-threatening when it occurs. The present report aimed to present a case in which a fatally idiopathic spontaneous hemoperitoneum in a 50-year-old male with bilateral facet subluxation at the C5-6 spine, including incomplete cord lesion (American-Spinal-Injury-Association: grade-C) and without associated injury, caused by falling from a 3-meters-scaffold.

To the best of the authors' to-date knowledge, the case in the present paper is the first report of idiopathic spontaneous hemoperitoneum in a patient with associated orthopedic injury. A review of the update literature was also carried out.

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Case Report

A 50-year-old man, construction worker (illegal immigrant), fell from a 3-meters-scaffold at the

construction area. He was immediately transferred to the emergency department at our center. On initial survey, the patient was alert, complaining of severe neck pain. He also had weakness and numbness in both upper and lower extremities. The vital signs were stable. The physical examination revealed motor grade III of deltoid and biceps muscle and grade I of myotome below this level, according to muscle grade criteria[11]. There was impaired sensibility of the skin as below C6-dermatome. The rectal examination revealed normal perianal sensation and sphincter tone. The bulbocavernosus reflex was positive. The plantar reflexes were in extension response. Additional examination showed no associated injuries. The physical examination for other systems according to Advanced Trauma Life Support guideline (ATLS), including head, chest, abdomen, pelvis, and extremities, were intact. The evaluation was completed by co-operation of multidisciplinary specialists as orthopedic surgeons, traumatologist, and neurosurgeon. The Focused Assessment with Sonography in Trauma (FAST) were serially performed and revealed normal findings.

The radiographs of cervical spine revealed blunting of the anterosuperior vertebral body margin of C6 and bilateral facet dislocations as 50% translation of upper vertebral body of C5 on C6 as defined as distractive-flexion stage III according to Allen's classification[11] (Fig. 1). Initial pelvic and chest radiographs were unremarkable. The initial management was reduction with skull traction under close observation. Reduction was engaged and maintained with 20 lbs traction without addition neurological deficit (Fig. 2). He also obtained intravenous methylprednisolone for 24 hours according to National Acute Spinal Cord Injury Study III (NASCIS-III) guideline[11].

The magnetic resonance imaging (MRI) for cervical spine was obtained subsequently (Fig. 3). It depicted the increased signal intensity of inter-vertebral disc at C5-6 level with discontinuity of posterior longitudinal ligament at this level. The posterior disc herniation was seen at central region of C5-6, C6-7 level with anterior and posterior indentation of central cord. There was also the disruption of ligamentum flavum at C5-6 level with the encroachment of lamina causing spinal canal narrowing. In addition, increased signal intensity of spinal cord at C5-6 level was reflecting traumatic myelopathy (Fig. 4).

For the definitive management, the operation was indicated for neural decompression and

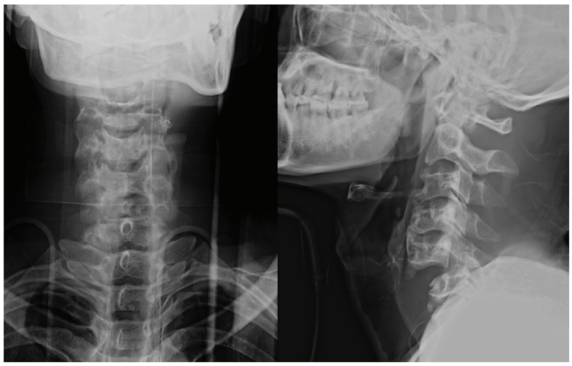

Fig. 1 Anteroposterior and lateral radiograph of the cervical spine show bilateral facet dislocations of C5-6 spine

Fig. 2 Lateral radiograph of the cervical spine reveals an in-line configuration after maintenance of reduction at 20 lbs

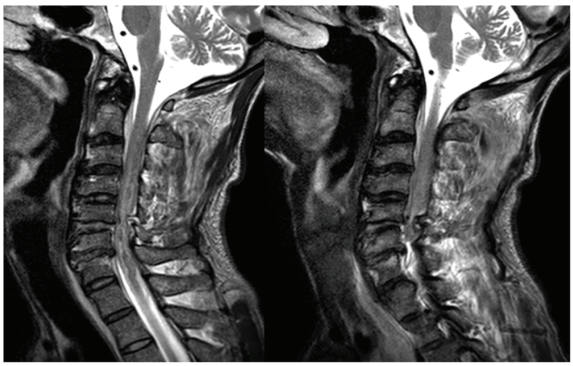

Fig. 3 Sagittal T2-weighted MRI shows the involvement of spinal cord at C5-6, C6-7 level

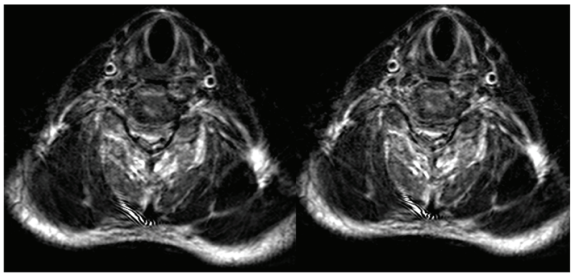

Fig. 4 Axial T2-weighted MRI demonstrates markedly compression of C5-6 level

Fig. 5 Lateral radiograph of the cervical spine demonstrates satisfactory alignment after the operation

stabilization of the C5-6 level, which showed markedly involvement in this patient. The pre-operative evaluation was completed by co-operation of multidisciplinary specialists as orthopedic surgeons, traumatologist, anesthesiologist, neurosurgeon, neurologist, and internist. His blood test revealed white blood cell count of 12,300/mm3, hematocrit of 35.3%, serum hemoglobin concentration of 12.1 g/dl, with normal platelet level (269,000/mm^3), blood urea of 12.5 mg/dl, and creatinine level of 1 mg/dl. The laboratory data of chemistry and other findings were within normal limits. His chest and pelvic radiographs were normal. The electrocardiographic findings were also normal. After the completion of the previous process, he underwent the anterior cervical discectomy, fusion with iliac bone graft, and plating of the C5-6 level at 20 hours after an injury (Fig. 5).

After the operation, this patient was in routinely postoperative protocol. He was on Philadelphia collar and started rehabilitation as early ambulation in gradually upright position. There was no additional neurological deficit as compared with preoperative status. Chest physical therapy was also begun with other programs. An endotracheal tube was removed at postoperative day 3. However, he had presented with right lung atelectesis and pneumonia. The sputum cultures were positive with *Pseudomonas aeruginosa* and Methicillin-sensitive *Staphylococcus aureus*. He was treated and recovered with intravenous ceftazidime and meropenam for a week. Other problems were hyponatremia and strongiloidosis from his feces. These points were treated by electrolyte correction and oral albendazole respectively. His ambulatory ability improved gradually. Previous medical problems were totally solved. At postoperative day 14, he was able to ambulate as bedside sitting with partial support. At this duration, he was planned to be discharged and continue further rehabilitation with home programs. However, during regular monitoring every two hours, he developed acute hemodynamic instability and loss of consciousness at postoperative day 15. Body temperature was 36.5°C, blood pressure 92/61 mmHg, pulse rate 94/min and respiratory rate 24/min. His chest radiographs was normal. The electrocardiographic findings were also normal. The source of effective volume loss was not found. He was strenuously resuscitated with intravenous crystalloid. Blood test was sent immediately. His blood test showed white blood cell count of 14,300/mm^3, a hematocrit of 31.1%, serum hemoglobin concentration of 11 g/dl, with a normal blood platelet level (368,000/mm^3), a blood urea of 21 mg/dl, and a creatinine level of 0.5 mg/dl. Laboratory data of chemistry and other findings were within normal limits. During the aggressive resuscitation, cardiac arrest occurred in 30 minutes since shock been diagnosed. Cardiopulmonary resuscitation (CPR) was implemented according to Advance Cardiac Life Support guideline (ACLS). CPR was performed 45 minutes and finished due to no signs of his recovery. This patient was sent to the Department of Forensic Medicine shortly after his death. Postmortem examination was performed.

Autopsy findings

Postmortem examination showed free intra-peritoneal blood 1,000 ml and fresh blood in gastric lumen-small bowels 500 ml without the identification of definitive source of bleeding, including vessel

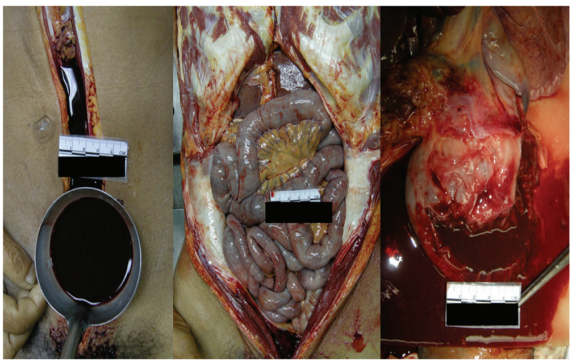

Fig. 6 The postmortem examination revealed fresh blood in the intra-peritoneal cavity and the gastric lumen

ruptures, mesenteric abnormality, gastritis or duodinitis, and gastric or duodenal ulcers (Fig. 6). There was no rupture point of the hollow-viscus organ. The solid organs were also intact, including the cerebrovascular and cardiopulmonary systems.

Discussion

Although spontaneous intra-abdominal bleeding is relatively rare, its reports could be fairly found. The previous literatures reported that this condition frequently related with pregnancy, gynecologic problems, and rupture of hepatocellular carcinoma (HCC)[1,2,6-8,10]. In addition, this condition was also reported in the association with coagulopathy, vascular, biliary, gastric, colonic, pancreatic, splenic, and other hepatic causes[3-5,9,12,13]. Vivarelli et al reported a retrospective review of 518 patients with HCC in their center during 1982-1993[14]. They revealed that approximately 2% of these patients presented with spontaneous rupture of their tumors. Other causes of spontaneous hemoperitoneum of in-hospital patients were ruptured ovarian cyst (44%), acute pancreatitis (9%), ruptured abdominal aortic aneurysm (6%), necrotizing gall bladder (3%), ruptured liver (in cases of eclampsia) (3%), ruptured spleen (3%), necrotizing uterine tube (3%), uterine cancer (3%), ectopic pregnancy (3%) and rarely metastatic disease such as gestational trophoblastic disease[14]. However, idiopathic spontaneous intra-peritoneal hemorrhage is actually extraordinary. From the best of the authors' to-date knowledge, the authors' current case was the first report of the idiopathic spontaneous hemoperitoneum in a patient with associated orthopedic injury.

Fewer case reports of idiopathic spontaneous hemoperitoneum have been proposed. Those reports explained that although they were performing a thorough exploration laparotomy, bleeding site frequently remained unclear in some cases[15-17]. In most of the reported cases, sources of bleeding were still unidentified[18]. As in the present case, there was no obvious source of bleeding in autopsy findings. Although there was fresh blood in gastric lumen-small bowels 500 ml, the forensic doctor still confirmed that there was no definitive source of this bleeding. Nishiyama et al reported an autopsy case of unexpected sudden death due to spontaneous "double-rupture phenomenon" of the gastric dissecting aneurysm[19]. It may relate to this phenomenon in a case of the present report with the bleeding in both gastric-duodenal lumens and intraperitoneal cavity. However, the forensic doctor could not confirm these findings as the previously mentioned discussion.

From the literature review, most cases of spontaneous hemoperitoneum frequently presented with acute abdominal pain. Patients might present with a wide variety of clinical presentations[16,20]. Physicians will consider hemoperitoneum in provisional diagnosis of patients who have a previous injury or history of anticoagulants use or concurrently abdominal tumor when they display clinical presentation of hypovolemia, or have decreasing hematocrit[10,18]. As previously mentioned, however, idiopathic event of hemoperitoneum is rare, and as result of poorly realized, which may lead to hazardous diagnostic postponement. Some patients had substantial hemoperitoneum without an obviously preceding symptom[15,20,21]. Therefore, it was sometimes difficult to diagnose before abdominal exploration. Some reports proposed the advantages of various imaging methods for early recognition of this condition[7,21]. In the present report, the presented patient had no complaint of preceding abdominal symptoms in both pre-and-postoperative periods. His visceral sensibility might be loss, due to incomplete cord lesion, as it was according to the absence of his sensation along with the dermatomes below the level of cervical spine injury. At this point, he might have an incompetency of detection of intra-abdominal abnormality when it occurred. Thus, this patient had no classic symptoms of spontaneous hemoperitoneum. This made the diagnosis of this condition quite difficult in the presented patient. This situation was very uncommon and essentially unique.

Conclusion

The authors recommend "a high index of suspicion" of spontaneous hemoperitoneum as the

possible cause of acute hemodynamic instability in patients with neurological deficit, due to cervical spine injury or any cause, which might obscure them to detect their antecedent abdominal pain. Meticulously physical examination in a patient with associated neurological deficit might assist to identify spontaneous intra-abdominal bleeding earlier that lead to rapidly perform the proper management.

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ภาวะตกเลือดในช่องท้องที่เกิดขึ้นเองและทำให้เสียชีวิตในผู้ป่วยที่มีการบาดเจ็บของกระดูกสันหลังบริเวณคอ

ชญานิน อ่างทอง, ธเนศ วรรธนอภิสิทธิ์, ชลเวช ชวศิริ

จุดประสงค์: *ภาวะตกเลือดในช่องท้องที่เกิดขึ้นเองเป็นภาวะที่พบน้อยมากและยังไม่มีการรายงานถึงกรณีดังกล่าวในผู้ป่วยที่มีการบาดเจ็บร่วมทางออร์โธปิดิกส์ การศึกษานี้ได้รายงานกรณีศึกษานี้ในผู้ป่วยที่มีการบาดเจ็บของกระดูกสันหลังบริเวณคอจากอุบัติเหตุ*

รายงานผู้ป่วย: *ผู้ป่วยชายไทยอายุ 50 ปี ได้รับการนำส่งโรงพยาบาลหลังจากตกนั่งร้านสูง 3 เมตร ผู้ป่วยได้รับการวินิจฉัยว่ามีกระดูกคอเลื่อน (bilateral facet subluxation of C5-6 spine) ร่วมกับภาวะไขสันหลังบาดเจ็บแบบไม่สมบูรณ์จากนั้นผู้ป่วยได้รับการผ่าตัดรักษาบริเวณคอ (anterior cervical discectomy and fusion) หลังผ่าตัดผู้ป่วยฟื้นฟูโดยรวมดีขึ้นตามลำดับแม้ว่าภาวะไขสันหลังบาดเจ็บแบบไม่สมบูรณ์นั้นยังคงที่ เมื่อวันที่ 15 หลังผ่าตัดผู้ป่วยมีสัญญาณชีพแย่ลงอย่างรวดเร็วและเสียชีวิตโดยไม่มีอาการนำมาก่อน การชันสูตรพบเลือดสดในช่องท้องปริมาณ 1,000 มิลลิลิตร และในกระเพาะอาหารรวมทั้งลำไส้เล็กปริมาณ 500 มิลลิลิตร โดยไม่พบจุดที่เป็นสาเหตุของการตกเลือดที่ชัดเจน*

การอภิปราย: *ผู้ป่วยรายนี้ไม่มีอาการผิดปกติชัดเจนนำมาก่อนการรับรู้ความผิดปกติของอวัยวะภายในช่องท้องอาจถูกบดบังจากภาวะไขสันหลังบาดเจ็บ ทำให้ผู้ป่วยไม่มีอาการแสดงที่พบได้เช่นในภาวะตกเลือดในช่องท้องทั่วไป การวินิจฉัยจึงกระทำได้ยากในผู้ป่วยรายนี้*

สรุป: *ภาวะตกเลือดในช่องท้องที่เกิดขึ้นเองเป็นภาวะหนึ่งที่ควรได้รับการพิจารณาว่าอาจเป็นสาเหตุของภาวะช็อกอย่างเฉียบพลันในผู้ป่วยที่มีการรับรู้ของเส้นประสาทลดลง จากกระดูกคอและไขสันหลังบาดเจ็บซึ่งการรับรู้ที่ลดลงนั้นทำให้ผู้ป่วยไม่สามารถตรวจจับอาการผิดปกติของช่องท้องและแสดงอาการออกมาตั้งแต่เริ่มแรกได้*