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Case Report

Steroid response in a rare case of vertebrae butterfly with spinal cord compression caused by chronic subdural hematoma

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Abstract

Introduction: Spinal subdural hematoma (SSH) is a rare entity. Clinical symptoms include motor, sensory and/or autonomic dysfunction due to compression of the spinal cord. There is no standard consensus in the management of SSH. Here, we report a rare case of SSH of sudden onset following trauma, successfully treated with conservative therapy.

Aim: This case highlights butterfly vertebrae as an incidental imaging finding before SSH diagnosis.

Case study: A 30-year-old female presented with weakness in the lower extremities after trauma, accompanied by paresthesia, urinary retention, and impaired defecating function. The thermoregulatory sweat exam revealed sudomotor dysfunction. Multiple slice computed tomography (MSCT) examination of the whole spine revealed wedge deformity of vertebral corpus (VC) L4 on the right lateral side, and VC L5 butterfly vertebrae. Spine magnetic resonance imaging (MRI) revealed an extramedullary intradural lesion at the T7 to T10 level suggestive of subdural hematoma.

Results and discussion: The administration of methylprednisolone pulse therapy, combined with physical rehabilitation, led to a rapid and significant improvement in the patient's motor function. The absence of the spinal pain possibly indicated that there was no more extension of the hematoma; motor recovery, even if slight, should be a predictor of successful conservative therapy, and physical rehabilitation.

Conclusions: SSH is an uncommon condition, with varying clinical presentation, and progression. This case report highlights significant improvement following conservative steroid treatment of a SSH. It also highlights a unique incidental finding of butterfly vertebra in a patient with paraparesis following trauma.

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1. INTRODUCTION

Spinal subdural hematoma (SSH) is of rare occurrence, and its pathological abnormalities may implicate in significant neurological dysfunction with profound morbidity and mortality. The incidence is higher in women, in individuals aged 50–70 years, and often involves the thoracic and thoracolumbar regions. Clinical symptoms of SSH of the spinal cord include motor, sensory and/or autonomic dysfunction due to spinal cord compression.[1]

SSH is often caused by trauma, lumbar puncture, and spinal surgery, but can also occur spontaneously in patients with coagulation disorders, vascular malformations, and neoplasms. Based on etiology, the most common causes include blood dyscrasias (32%), anticoagulant use (16%), major trauma (18%), minor trauma (16%), lumbar puncture (14%), vascular malformations (4%), and spinal surgery (4%).[2,3] Since the condition is a rare entity there is currently no consensus on the ideal management of SSH.

2. AIM

In this paper we present a rare case of SSH of sudden onset, successfully treated with conservative therapy. The case also highlights an incidental finding of butterfly vertebrae, which although did not particularly contribute to the chief complaint, is an interesting imaging finding in the patient prior to discovering the SSH.

3. CASE STUDY

A 30-year-old female presented with a chief complaint of weakness in both legs after falling from a height of 3 m 9 days prior, in addition to lower back pain accompanied by tingling and numbness, radiating to the feet. She also complained of difficulties in urinating and defecating immediately after the incident. The patient's initial numerical rating scale (NRS) for pain was 5–6, while all other vital signs were within normal limits.[4] The patient's medical history reveals no prior conditions related to blood disorders, and there is no history of anticoagulant medication use. On neurological examination, she showed decreased movement in the lower extremities (particularly in the gluteus maximus and iliopsoas muscles), with a bilateral motor strength score of 4 and increased deep tendon reflexes (knee pes reflex – KPR and achilles pes reflex – APR; grade +3), accompanied with the presence of pathological Babinsky reflex bilaterally. The sensory exam showed hypesthesia from the foot to the level of the L1 and L2 dermatomes. Impaired autonomic function (urinary and fecal retention) was observed. On examination of the vertebral column, there was no visible injury or hematoma, although tenderness and knocking pain were present at the lumbar vertebral corpus (VC) L4 and L5, and the range of movement (ROM) was limited due to pain. The Laseque's, Patrick and counter Patrick tests were positive bilaterally. Based in these findings the patient was classified as incomplete (Type D) category of the American Spinal Injury Association (ASIA) impairment scale. Pelvic and lumbosacral X-ray scans revealed a wedge deformity of the lumbar VC L4 on the right lateral side, and deformity of VC L5. The thermoregulatory sweat exam revealed sudomotor dysfunction, indicated by the lack of color change from soles of the foot until the level of T8 and T9 dermatomes.[5] The multiple slice computed tomography (MSCT) examination of the whole spine revealed wedge deformity of VC L4 on the right lateral side, and VC L5 butterfly vertebrae (Figure 1).

A non-contrast magnetic resonance imaging (MRI) examination of the whole spine revealed multi-focal extramedullary intradural lesions at the VC T7 to T10 level suggestive of SSH. In the following week (approximately day 15 after onset), a contrast whole spine MRI was performed, confirming no visible intramedullary/extramedullary intradural vascular lesions or extradural lesions (Figure 2).

The patient received intravenous methylprednisolone therapy for 12 days starting from day 2 until day 14 after onset, with a starting dose of 125 mg every 8 h (gradual tapering down every 3 days), and was the started on a rehabilitation program on day 16 after onset. The follow up on day 21 after onset revealed improved motor strength (from strength level 4 to 5)[6] and autonomic function (the patient was able to regain control of urinating and defecating function). There was improvement of tendon reflexes and lack of pathological reflex. Likewise, the patient also started to regain sensory function, and reported no more pain.

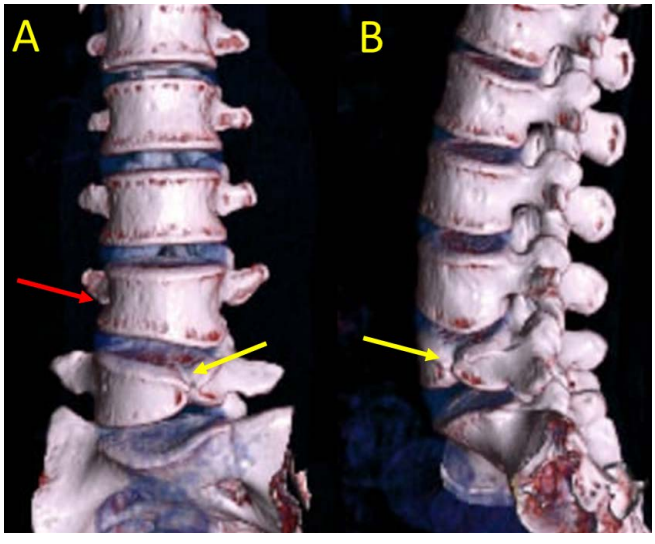

Figure 1. MSCT scan of the lumbar vertebrae. Comments: (A) anterior-posterior view showing wedge deformity of VC L4 on the right lateral side (red arrow) and butterfly vertebra L5 (yellow arrow); (B) lateral view showing butterfly appearance of VC L5 from the lateral side.

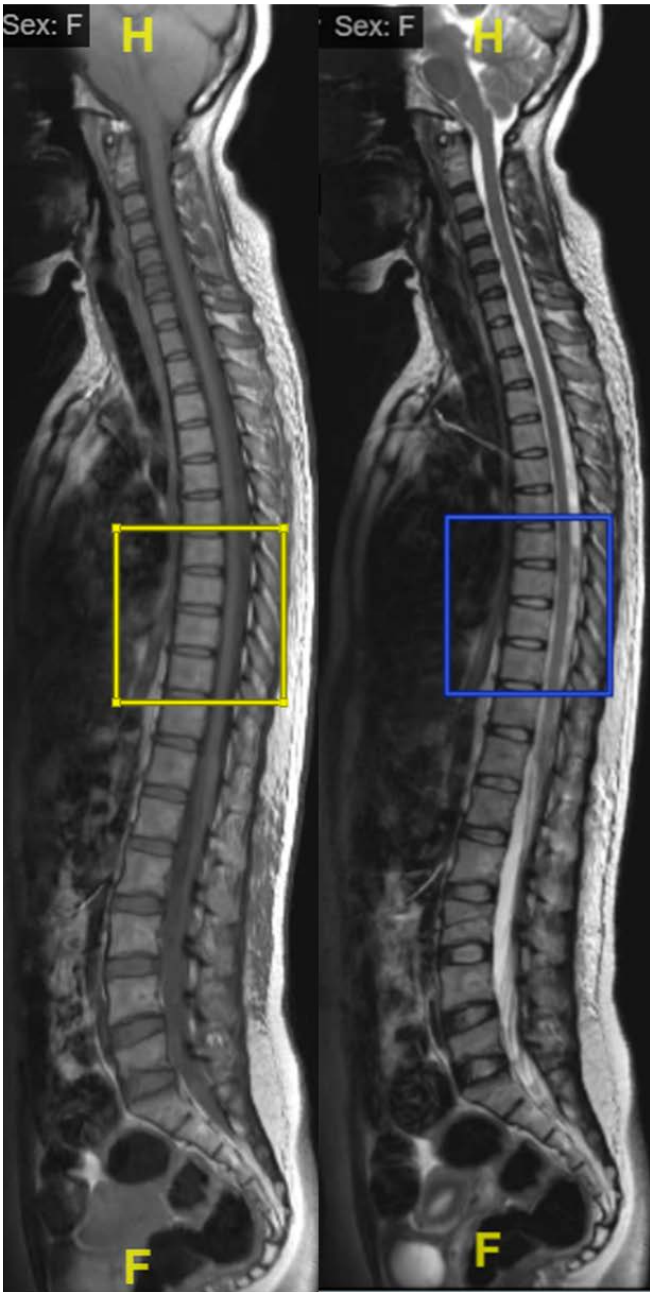

Figure 2. Non-contrast MRI of the whole spine showed multi-focal isointense lesions on T1W and hypointense lesions on T2W in the extramedullary intradural space at levels T7 to T10, suggestive of a subdural hematoma. Comments: yellow box – T1W, blue box – T2W.

4. DISCUSSION

Based on the mechanism, SSH are divided into 3 categories including those associated with bleeding (patients on anticoagulants, hemophilia patients), major trauma (presence of fracture), or minor trauma (no signs of fracture, for example due to lumbar puncture).[7] In this case, the patient was classified in the category of hematoma associated with minor trauma because there were no signs of fracture in the region of hematoma. Meanwhile, the VC L5 butterfly vertebrae found in the patient is likely a congenital anomaly that occurred due to failure of fusion of two chondrifications. The butterfly vertebrae are typically rare, asymptomatic, and usually an incidental finding when a spinal cord injury occurs. In cases of symptomatic butterfly vertebra, clinical signs and symptoms can be related to other functional disorders, 27% of which present as decreased reflexes, L5 weakness, sensory deficits, sciatica, and spasticity.[8,9]

SSH can also be classified based on onset and clinical progression. It is deemed acute when onset is between hours-days, marked by sudden onset of back pain followed by radicular pain and progressive paraplegia. The cause (trauma, lumbar puncture, coagulopathy) is usually obvious. In subacute SSH (a few days – 2 weeks), there is a more gradual onset of less well-localized pain in the back, hip, and leg, often accompanied by headache, followed by the development of weakness and numbness of the legs, which may progress to paraplegia over approximately 2 weeks. In chronic cases (2 weeks – years), signs and symptoms of progressive spinal cord compression occur over a period of months or years. Pain is either moderate or absent, accompanied with fluctuating weakness, spasticity, autonomic dysfunction (micturition and defecation) within said period.[2,10] In this case report, the SSH is classified as acute-subacute. This can also be supported by the results of a MRI spine, which is the main choice for diagnostics. The evolution of the intensity of hematoma on MRI can provide information about the duration of the SSH and also applies to hematomas that occur in the brain. In the first few days, isointense or hypointense lesion is found on T1-weighted (T1W) and T2-weighted (T2W) sequences. This is because the red blood cells (RBC) are still intact and the concentration of deoxyhemoglobin is high. After about 1 week, hyperintensity will appear, first seen on T1W due to the formation of intracellular methemoglobin, then followed on T2W due to formation of extracellular methemoglobin followed by RBC lysis. In the chronic phase, diffuse hyperintensity will be seen in the hematoma region.[2]

The mechanism of SSH is not fully known, and differs to the formation of intracranial subdural hematoma. The first theory is that SSH does not occur from direct trauma and tearing of bridging vessels, but instead due to a rapid increase in intraspinal segment pressure in the lateral spinal arteries and veins. This pressure increase cannot be neutralized by increasing spinal fluid pressure due to the protective effect (shielding effect) of the spinal column and its ligaments. The second theory is that the subdural space, situated between the arachnoid and dura mater, is connected by a weak and easily torn cell boundary layer. Traumatic tearing of the dura layer results in an open gap in the subdural space. The third, is that blood produced can enter the subdural space due to rupture of the radicular arteries along nerve roots or due to damage to the epidural Batson plexus, causing fluid withdrawal through negative osmotic pressure and hydrostatic pressure gradients, resulting in fluid extravasation into the subdural space. Leakage of blood and fluid into the subdural space can be aggravated by disrupt-

ing intrabdominal and intrathoracic pressure which can cause blood vessel rupture.[2,11,12]

Clinical symptoms include spinal pain, radiculopathy, progression of paraparesis/paraplegia, and other neurological deficits, depending on the location and severity of the nerve compression. It is stated that a pressure of 300 g/cm^2 in a segment can cause real but reversible paraplegia, whilst a pressure increase (400 g/cm^2) results in irreversible paraplegia which can potentially leave residual or permanent symptoms.[9] In this case there was improvement in paraparesis, sensory deficits, and autonomic function without leaving residual symptoms, in other words the pressure inflicted by the SSH towards the spinal cord was 300 g/cm^2.

There is no standard consensus for the management of SSH, and approaches include conservative treatment, and surgical therapy. Surgical removal of a hematoma is typically indicated in the presence of progressive neurological deficits, although it does not always lead to favorable outcome, since results depend on hematoma size, degree of paralysis, progression, and timing of surgery.

In this case report, conservative therapy was carried out with good outcome, with the patient showing improved neurologic function. The patient also stated pain improvement. Hence, the absence of pain, in addition to improved motor, sensory, and autonomic function, are indications that there was no more extension of the hematoma, and is a sign of successful conservative therapy.[1] Several other case reports found favorable outcomes with steroid administration. In a case report by Song et al. (2011), the authors demonstrated that the administration of methylprednisolone is effective as a conservative treatment for SSH. In this report, patients received 1 gram of intravenous methylprednisolone daily for five days, followed by a regimen of oral prednisolone at an initial dose of 60 mg per day, tapered to 20 mg per day over the subsequent two months. Methylprednisolone was shown to reduce astrocyte cell death and microglial activation, suppress A1 astrocyte activation, and promote improvements in nerve function.[1,3,13,14] Recently, Jashari et al. (2024) described a cohort of patients with a history of autoimmune thyroiditis who presented with neurological and psychiatric manifestations consistent with autoimmune encephalopathy. Following diagnosis, patients received methylprednisolone pulse therapy (1 g administered daily for 5 days). Subsequently, they were discharged on an oral corticosteroid regimen, which resulted in significant clinical improvements, evidenced by the resolution of seizure activity and a marked reduction in confusion.[15]

5. CONCLUSIONS

(1) SSH is an uncommon condition, with varying clinical presentation and progression, in accordance to the lesion location and the intensity of spinal cord compression.
(2) This case report highlights significant improvement following conservative steroid treatment and physical rehabilitation of a SSH.
(3) It also highlights a unique incidental finding of butterfly vertebrae in a patient with paraparesis following trauma.

Conflict of interest

None.

Funding

None.

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Ethics

Ethics approval and consent to participate. This study was approved by the ethical committee at the Health Research Ethics Committee, Faculty of Medicine, Hasanuddin University, Makassar. All patients in this case report have provided written informed consent regarding the writing and publication of the report.

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