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Spontaneous Cervical Epidural Hematoma Following Anti-Coagulant Medications with Quadriparesis: A Case Report and Narrative Review

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Abstract

Spontaneous spinal epidural hematoma is an uncommon entity. We present a 42-year gentleman who was a known case of chronic deep vein thrombosis of leg on Tab. Acitrom (6 mg OD) for one & half year presented with sudden-onset weakness of bilateral upper & lower limbs with bowel & bladder involvement. MRI showed ventral epidural haematoma extending from the lower margin of C2 to C6 with severe canal compromise. Patient underwent emergency decompression with hematoma evacuation on the same day within 06 hours of presentation to our centre. At 2-year follow-up patient has recovered good bowel and bladder control and has a residual left leg foot drop. Spontaneous spinal epidural hematoma is a rare condition where early management is the key. If associated with neurodeficits, early decompression (<48 hours) is indicated for better prognosis. This case report highlights the fact if dealt pro-actively patient can have a good neurologic recovery.

Keywords: Epidural hematoma, Quadriparesis, Cervical spine, Anti-coagulants

Introduction

Spinal hematoma was first described as "spinal apoplexy" by G.J. Duverney in 1682 in cadavers. But the clinical condition was first described by Jackson in 1869 [1]. The annual reported incidence is <1 in 100000 in general population. The importance of early clinical diagnosis & management lies in the fact – morbidity & mortality associated with the condition. In about 70% of the cases an underlying cause can be found which includes – iatrogenic (lumbar puncture), trauma, coagulopathy, arterio-venous malformation (AVM), tumour apoplexy & commonly post-medications [2].

Case report

We report a 42-year-old gentleman who is a known case of chronic deep vein thrombosis of left lower limb on Tab. Acitrom (Acenocoumarin) 6 mg OD (Once daily) who presented with sudden onset weakness of both upper & lower limb with bowel & bladder involvement. There was no history of fall or trauma (Figure 1, 2). He presented 24-hours after the onset of weakness to our centre. On examination the motor power in upper & lower limb was Grade 0 on MRC (Medical research council) scale. His score was 0/60. Lowest sensory dermatome was C5. His anal tone and bulbo-cavernous reflex were absent (AIS – ASIA Impairment Scale Grade A). MRI suggested an Isointense T1 & Hypointense T2 anterior cervical epidural collection from lower margin of C2 vertebra to mid-C6 vertebra (Figure 3, 4, 6). Finding of Isointense in T1 & Hyperintense in T2 suggested it is Acute stage (1-3 days) STIR & GRE imaging revealed severe cord compression at C4 & C5 with posterior displacement of cord. (Figure 5, 7) Cord oedema was extending up to C2 vertebra (Figures 4) [3]. Venous doppler of bilateral lower limb showed chronic deep vein thrombosis of left popliteal artery and subacute partial thrombosis of right superficial femoral artery. His coagulation profile showed his INR (International Normalized Ratio) 14.1 (Normal value 2-3), PT (Prothrombin time) 132.3 (Normal value – 10-13 seconds) & PTT (Partial thromboplastin test) 99.1. (Normal value – 25-35 seconds). He was found to be negative for SARS CoV-2 on PCR (Polymerase chain reaction). Haematologist was consulted and Fresh frozen plasma was started immediately. After cross-consultation with other specialities a decision was made to offer an option of surgical decompression to patient. Patient attenders were clearly counselled regarding prognosis. Initial surgical plan was emergent cervical spine decompression from posterior spinal exposure and Inferior vena cava filter in second stage. No steroids were administered pre-operatively [4].

Patient was planned for emergent decompression within 6 hours after presenting to our centre (30 hours from onset of weakness). Neuromonitoring could not be deployed in the about present scenario. Patient was put on Mayfield traction after turning prone

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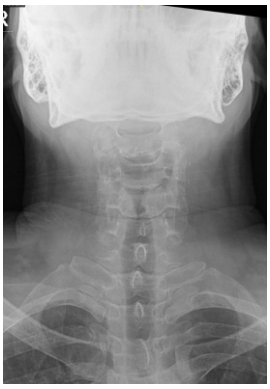

Figure 1: Pre-operative cervical spine Antero-posterior x-ray

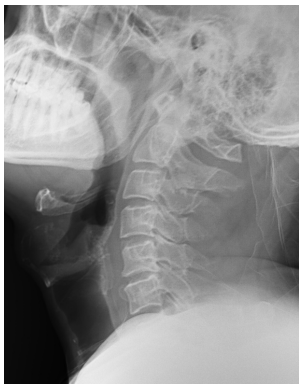

Figure 2: Pre-operative Lateral Cervical Spine X-ray

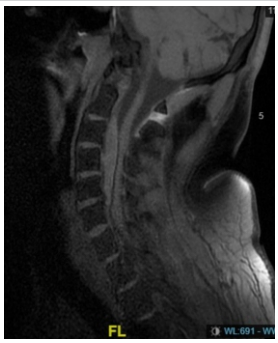

Figure 3: Pre-operative T1W Sagittal Image showing Hyperintense Epidural hematoma from lower margin of C2 vertebra to upper margin of C6 vertebra spanning at least 3.5 vertebra.

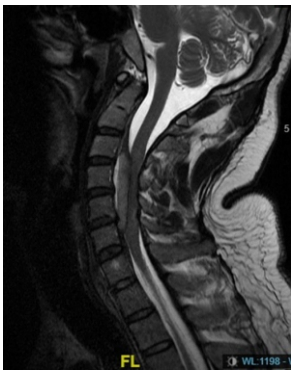

Figure 4: Sagittal T2W MRI image showing compression and spinal cord with cord oedema extending up to C2 vertebra.

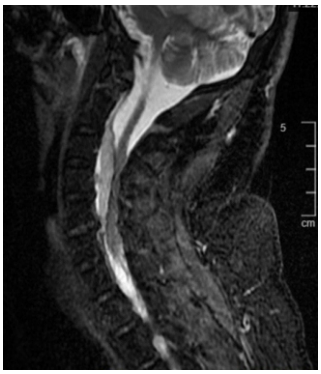

Figure 5: Sagittal STIR Image showing hematoma and amount of cord compression.

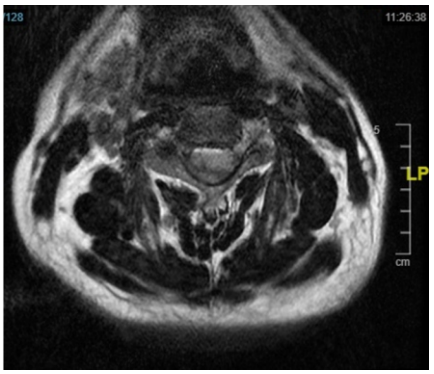

Figure 6: Pre-Operative T2W Axial image showing cord compression.

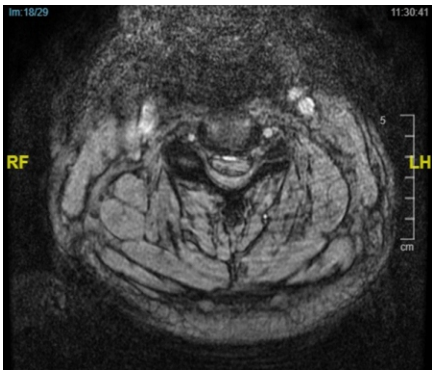

Figure 7: STIR Axial Image showing cord compression.

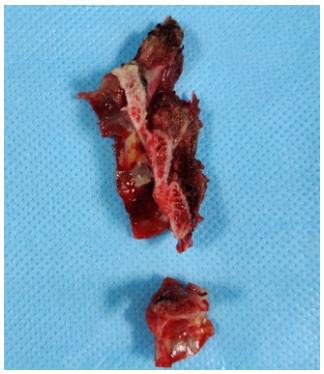

Figure 8: Lamina removed

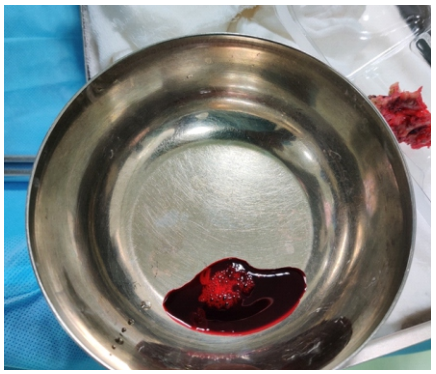

Figure 9: Hematoma aspirated.

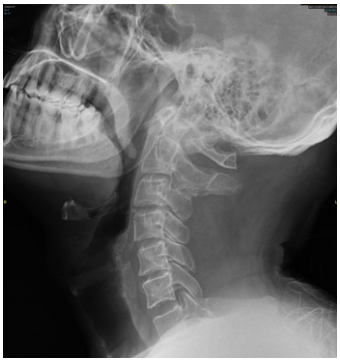

Figure 10: Post-operative x-rays showing antero-posterior view of cervical spine.

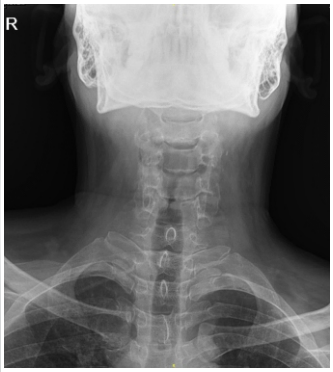

Figure 11: Post-operative x-ray showing laminectomy with no evidence of instability.

position under general anaesthesia. Posterior cervical spine exposure was done from C2 vertebra to C7 vertebra after confirming levels by intra-operative palpation and image-intensifier. Cervical laminectomy was done from C3 to C6 by using a high-speed burr (Aesculap Inc., PA, U.S.A.) to make paravertebral gutters at spinolaminar junction and the remaining part of the bone removed by rongeurs (Figure 8). Adequate cord compression was obtained after laminectomy and cord was gently retracted and epidural blood aspirated. About 10 ml of blood was aspirated (Figure 9). Further exploration of the hematoma was curtailed by the epidural bleeding. Since the blood coagulation profile was deranged, intra-operative transfusion of about 6 Fresh frozen plasma & crystalloids was given by the anaesthetist. Wound was closed in layers after obtaining

Table 1: showing the coagulation profile from presentation to discharge.

	Prothrombin Time (Normal 10-13 seconds)	International Normalized Ratio (INR) (Normal 2-3)	Mean Normal Prothrombin Time (Test – 29.7 seconds)
Pre-operative	132.35 s	14.1	11.4
Post-Operative Day 1	16.4 s	1.5	11.4
Post-Operative Day 2	12 s	1.1	11.4
At Discharge	11.9 s	1.1	11.4

Table 2: showing motor power at final follow-up of 02 years

Power grade	Right	Left
C5, C6, C7	5-May	5-Apr
C8 & T1	5-Mar	5-Mar
L2, L3, L4	3+/5	5-Mar
L5 & S1	3+/5	0/5

adequate haemostasis over a suction drain. We did not put any epidural drainage. Post-operatively patient was on ventilator support and needed tracheostomy in view of right lower lobe consolidation of lung. Venogram revealed right external and common iliac vein severe stenosis. Due to above findings IVC filter was not performed. Post-operatively patient was on low molecular heparin & his coagulation profile on Post-operative Day 1 was as followed PT 16.4 (Normal value – 10-13 seconds), INR 1.5 (Normal value 2-3), MNPT –11.4 (Normal value – 20-40). At discharge anti-coagulant was changed to Tab. Dabigatran 75 mg BD (twice daily) (Table 1). On Post-Operative follow up at 6 weeks he had considerable improvement in bowel & bladder symptoms and was on clean intermittent self-catheterisation, his upper limb strength was 4/5 on right side & 3/5 on left side. He had symptoms of myelopathy hand. Lower limb was spastic, and power was fair. In next follow-up after 3 months from surgery - Bladder was on intermittent self-catheterization & Power was as follows: (Table 2)

At final follow-up he has improved considerable neurologically and is able to ambulate with a single elbow crutch, has good bowel & bladder control and is using a foot drop splint on left. (AIS Grade D). No evidence of instability was noted on follow-up x-rays (Figure 10, 11).

Discussion

In the first case report published by Jackson in 1869, a fourteen-year-old girl started developing weakness in hands and after 3 days of initial onset of symptoms, weakness in respiratory muscles noted leading to respiratory arrest & death. A clinical pointer towards epidural hematoma is rapidity with which symptoms develop which differentiates it from other causes of compressive myelopathy. In our case it was sudden onset weakness and given the fact that patient was on anti-coagulant, it proved to be the putative cause. Predisposing factors include anticoagulant therapy for prosthetic cardiac valves, deep vein thrombosis, therapeutic thrombolysis for acute myocardial infarction, haemophilia, factor XI deficiency, long term aspirin use, cocaine abuse, vascular malformation, Paget's disease, and pregnancy.

The term spontaneous means without any pre-existing trauma. Some authors also include the definition as "without any pre-existing aetiology" which excludes anti-coagulant therapy from word "spontaneous" [6]. But most other studies don't follow this definition and we would like to call it as Spontaneous Spinal Epidural Hematoma (SSEH) or Spontaneous Cervical Epidural Hematoma (SCEH).

In one of the largest reviews of spinal epidural hematoma Kreppel et.al states that anti-coagulant therapy alone doesn't cause hematoma & haemorrhage usually occurs at the site of least resistance with some additional trigger [2]. Beatty and Winston postulated that the source of bleeding for spinal epidural hematomas was the free anastomotic arteries that run in the epidural space and connect with radicular arteries rather than venous plexus. One of the reasons given is that the pressure in venous system is not "enough" to cause cord compression. The rapidity with which the myelopathy follows is like AVM (Arterio-venous malformation) in which case the pressure is like arterial system [5]. We believe in our case given the location and compression - arterial bleeding may have been the source.

With regards to location in spine, it can be epidural, subdural, subarachnoid, or intramedullary. Location can be dorsal, dorsolateral, or ventral in epidural space [3]. Most commonly hematoma occur in dorsal location at cervicothoracic and thoracolumbar regions. Postulated reason being a junctional region can have abnormal mobility and presence of Hoffmann ligaments connecting dura to posterior longitudinal ligaments [7]. In our case hematoma was ventral. Acute cervical epidural hematoma is fatal unless surgically evacuated. Since it is difficult to evacuate from anterior route which would lead to multiple corpectomies, we choose the posterior route for decompression. Also, posterior exposure is extensile & simple compared with anterior and fraught with less approach related complications.

Hematoma in epidural space is usually limited in number of vertebrae spanned which is in contrast with subarachnoid hematoma which is extensive. Lee HH et.al reported a case in which hematoma extending from C1 to sacrum [8]. In this case, authors have decompressed at the site of maximum decompression and used epidural drainage at above & below the site with a very good neurological recovery. In such a scenario non-surgical management is another option as highlighted by Raack et.al [9]. They state that if INR is medically correctable in an extensive hematoma and if the patient is high-risk and if there is early and sustained recovery then SSEH can be managed medically with acceptable outcomes, close monitoring is the key in such a scenario.

Symptoms of hematoma usually begin in radicular fashion and can progress either acutely in matter of hours or day or can lead to chronic course. Pain can be intense with knife-like pain i.e., stabbing character at the location of haemorrhage. ("coup de poignard"). Some cases can resemble acutely ruptured disc, epidural neoplastic condition, transverse myelitis, dissecting aortic aneurysm, congenital cysts, or abscess in epidural region. Rarely it can even present as Horner's syndrome or Brown-Sequard Syndrome [8].

Preoperative neurologic deficit is the main prognostic indicator, where the outcome is favourable for those with an incomplete preoperative sensorimotor deficit. In 49% of cases at least 04 vertebra were spanned. In our case it spanned from lower margin of C2 up to mid C6 (about 3.5 vertebra). Longer the hematoma more is the morbidity and poorer the prognosis. In a study of 30 consecutive

patients treated at a single centre Zhong et.al found that prognosis is better if it involved <4 vertebral segments, thoraco-lumbar or lumbar segments. Other prognostic markers include shorter progression interval, neurodeficits & spinal cord oedema – all led to poorer outcome [10]. Groen and Ponssen reported significantly better outcomes for patients with complete neurologic deficits who underwent decompression within 36 hr of symptom onset; for those with incomplete deficits, decompression was successful if performed within 48 hr of presentation [11]. In our case patient underwent decompression in less than 36 hrs after onset of symptoms.

Agnetti et.al first reported spinal epidural hematoma in an ankylosing spondylitis patient who developed generalised tonic-clonic seizures [12]. In a recent radiographic study Vierunen et.al reported the association of epidural hematoma in post-traumatic ankylosing spondylitis patients to be about 68% [13]. In a recent single institution retrospective review by Hanna et.al used deep 6 AI (Artificial Intelligence) platforms and identified 164 patients from 55 papers of ankylosing spondylitis with trauma, of which 17 had epidural hematoma. 14 were males and 3 were females, ranging from 51-88 years, with majority occurring at cervico-thoracic or thoraco-lumbar junction. All required surgery, 64.7% required decompressive procedures in addition to fixation of fracture and most improved neurologically after intervention. Thus, in a traumatic ankylosing spondylitis case with deteriorating neurology, having a differential diagnosis of epidural hematoma is important [14].

Liao et.al used modified Rankin Scale (mRS) to record functional recovery following SSEH (Table 3). In their study a score of 0, 1 or 2 were considered to have made a functional recovery [15]. Our case made a good recovery and was ambulant with an elbow crutch and was continent with regards to bowel & bladder control.

Table 3: Modified Raskin Scale (mRS)

Score	Description
0	No symptoms
1	No significant disability despite symptoms: able to perform all usual activities & duties
2	Slight disability: unable to perform all previous activities; able to care for own affairs w/o assistance
3	Moderate disability: requires some help, but able to walk w/o assistance
4	Moderately severe disability: unable to walk w/o assistance or attend to bodily needs.
5	Severe disability: bedridden, incontinent, & requires constant nursing care & attention

Conclusion

Spontaneous spinal epidural hematoma though a rare condition, clinician should know and suspect it in case of sudden onset deficit with no triggers. Given its myriad of symptoms in some cases having a good knowledge about the condition is important for all spine care specialist. As in all acute compressive myelopathy, early decompression is the key. This case report reiterates that early diagnosis and emergent intervention is the paramount for acceptable and safe outcomes.

Declaration of patient consent : The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his/her consent for his/her images and other clinical information to be reported in the Journal. The patient understands that his/her name and initials will not be published, and due efforts will be made to conceal his/her identity, but anonymity cannot be guaranteed.

Conflict of Interest: None; **Source of Support:** None

References

[1] Jackson R. CASE OF SPINAL APOPLEXY. The Lancet. 1869 Jul 3;94(2392):5–6.

[2] Kreppel D, Antoniadis G, Seeling W. Spinal hematoma: a literature survey with meta-analysis of 613 patients. Neurosurg Rev. 2003 Jan;26(1):1–49.

[3] MR imaging of spinal haematoma: a pictorial review - PMC [Internet]. [cited 2023 Mar 7]. Available from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC6541191/

[4] Hadley MN, Walters BC. Introduction to the Guidelines for the Management of Acute Cervical Spine and Spinal Cord Injuries. Neurosurgery. 2013 Mar;72 Suppl 2:5–16.

[5] Beatty RM, Winston KR. Spontaneous cervical epidural hematoma. A consideration of etiology. J Neurosurg. 1984 Jul;61(1):143–8.

[6] Baeesa S, Jarzem P, Mansi M, Bokhari R, Bassi M. Spontaneous Spinal Epidural Hematoma: Correlation of Timing of Surgical Decompression and MRI Findings with Functional Neurological Outcome. World Neurosurg. 2019 Feb;122:e241–7.

[7] Gopalkrishnan CV, Dhakoji A, Nair S. Spontaneous cervical epidural hematoma of idiopathic etiology: Case report and review of literature. J Spinal Cord Med. 2012 Mar;35(2):113–7.

[8] Lee HH, Park SC, Kim Y, Ha YS. Spontaneous Spinal Epidural Hematoma on the Ventral Portion of Whole Spinal Canal: A Case Report. Korean J Spine. 2015 Sep;12(3):173–6.

[9] Raasck K, Khoury J, Aoude A, Abduljabbar F, Jarzem P. Nonsurgical management of an extensive spontaneous spinal epidural hematoma causing quadriplegia and respiratory distress in a choledocholithiasis patient. Medicine (Baltimore). 2017 Dec 22;96(51):e9368.

[10] Zhong W, Chen H, You C, Li J, Liu Y, Huang S. Spontaneous spinal epidural hematoma. J Clin Neurosci. 2011 Nov;18(11):1490–4.

[11] Groen RJ, Ponssen H. The spontaneous spinal epidural hematoma. A study of the etiology. J Neurol Sci. 1990 Sep;98(2–3):121–38.

[12] Agnetti V, Monaco F, Mutani R. Post-convulsive spinal epidural

haematoma in ankylosing spondylitis. Eur Neurol. 1979;18(4):230–3.

[13] Vierunen RM, Koivikko MP, Siironen JO, Kerttula LI, Bensch FV. Post-traumatic spinal hematoma in ankylosing spondylitis. Emerg Radiol. 2021 Jun;28(3):601–11.

[14] Hanna G, Uddin SA, Trontis A, Ross L, Drazin D, Kim TT, et al. Epidural hematoma in patients with ankylosing spondylitis requiring surgical stabilization: a single-institution retrospective review with literature analysis. Neurosurg Focus. 2021 Oct;51(4):E5.

[15] 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 Jan;100(1 Suppl Spine):38–45.

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