Spontaneous Cervical Epidural Hematoma Following Anti-Coagulant Medications with Quadriplegia: 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, Quadriplegia, 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 facts – 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 bilateral upper & lower limbs 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].

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). Neuro-monitoring could not be deployed in the about present scenario. Patient was put on Mayfield traction after turning prone scenario. Patient was put on Mayfield traction after turning prone

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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

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.
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 catheterisation & Power was as follows: (Table 2) At final follow-up he has improved considerably 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 anti-coagulant 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

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

<table>
<thead>
<tr>
<th></th>
<th>Prothrombin Time (Normal 10-13 seconds)</th>
<th>International Normalized Ratio (INR) (Normal 2-3)</th>
<th>Mean Normal Prothrombin Time (Test – 29.7 seconds)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pre-operative</td>
<td>132.35 s</td>
<td>1.4</td>
<td>11.4</td>
</tr>
<tr>
<td>Post-Operative Day 1</td>
<td>16.4 s</td>
<td>1.5</td>
<td>11.4</td>
</tr>
<tr>
<td>Post-Operative Day 2</td>
<td>12.5 s</td>
<td>1.1</td>
<td>11.4</td>
</tr>
<tr>
<td>At Discharge</td>
<td>11.9 s</td>
<td>1.1</td>
<td>11.4</td>
</tr>
</tbody>
</table>

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

<table>
<thead>
<tr>
<th>Power grade</th>
<th>Right</th>
<th>Left</th>
</tr>
</thead>
<tbody>
<tr>
<td>C5, C6, C7</td>
<td>5-May</td>
<td>5-Apr</td>
</tr>
<tr>
<td>C8 &amp; T1</td>
<td>5-Mar</td>
<td>5-Mar</td>
</tr>
<tr>
<td>L2, L3, L4</td>
<td>3+/5</td>
<td>5-Mar</td>
</tr>
<tr>
<td>L5 &amp; S1</td>
<td>3+/5</td>
<td>0/5</td>
</tr>
</tbody>
</table>
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.

### 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.

### Table 3: Modified Raskin Scale (mRS)

<table>
<thead>
<tr>
<th>Score</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>0</td>
<td>No symptoms</td>
</tr>
<tr>
<td>1</td>
<td>Slight disability: unable to perform all previous activities; able to care for own affairs w/o assistance</td>
</tr>
<tr>
<td>2</td>
<td>Moderate disability: requires some help, but able to walk w/o assistance</td>
</tr>
<tr>
<td>3</td>
<td>Moderately severe disability: unable to walk w/o assistance or attend to bodily needs.</td>
</tr>
<tr>
<td>4</td>
<td>Severe disability: bedridden, incontinent, &amp; requires constant nursing care &amp; attention</td>
</tr>
</tbody>
</table>

### 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


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