CASO CLÍNICO/CASE REPORT

Spontaneous Cervical Epidural Hematoma

Hematoma Epidural Cervical Espontâneo

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Resumo

Introdução: O hematoma epidural espontâneo é uma entidade rara que pode levar a complicações graves se não for prontamente identificado. A realização de ressonância magnética é o melhor método de imagem para o seu diagnóstico.

Caso Clínico: Os autores reportam o caso de uma doente hipocoagulada, sem história prévia de traumatismo, que recorreu ao serviço de urgência por cervicalgia de início súbito, associada a parestesias de ambos os membros superiores, com evolução em algumas horas para tetraplegia e diminuição dos reflexos tendinosos profundos. A tomografia craniocefálica e cervical não mostrou alterações, tendo sido diagnosticado um hematoma epidural cervical por ressonância magnética. A doente apresentou evolução clínica favorável com tratamento conservador.

Conclusão: Este caso pretende demonstrar a complexidade diagnóstica dos hematomas epidurais e a possibilidade de sucesso terapêutico com uma abordagem conservadora, numa entidade em que o tratamento cirúrgico se mantém como o *gold standard*.

Abstract

Introduction: Spontaneous spinal epidural hematoma is a rare condition that leads to severe consequences if not promptly recognized. Magnetic resonance imaging is useful for a correct diagnosis.

Case Report: We report the case of a woman on anticoagulant therapy and no previous trauma that presented to the emergency department with sudden cervical pain and numbness of upper limbs which has evolved to quadriplegia with absent deep tendon reflex in a few hours. Head and cervical computed tomography were normal, but magnetic resonance imaging showed a cervical epidural hematoma. She had a good improvement with conservative treatment.

Conclusion: This case illustrates the complexity of the diagnosis of this entity and the possibility of successful treatment with a conservative approach, in a condition that surgery remains the gold standard treatment.

Introduction

Spontaneous spinal epidural hematoma (SSEH) is a rare condition, which can course with a prompt and progressive neurological deficit, requiring a timely diagnosis for a better outcome. Often described as a hematoma occurring in the absence of trauma or iatrogenic procedure,[1] spontaneous hematoma does not exclude causes such as coagulopathy, vascular malformations, cavernous angioma, and tumour.[2] Usually, it presents as a sudden neck pain, evolving to paraparesis or quadriparesis, depending on the level of the lesion.[3] The diagnostic method of choice is magnetic resonance imaging (MRI), and the gold standard treatment is surgery. Earlier interventions are associated with better outcomes.

Case Report

A 65-year-old female came to the emergency department (ED) due to a sudden cervicalgia associated to a slight limitation in the cervical movements and numbness of upper limbs, in the previous three hours. There was no known precipitating cause or relieve factors, including paracetamol.

In her medical history, she had paroxysmal atrial fibrillation (AF), arterial hypertension, dyslipidemia, depressive disorder, and a right ovarian cyst with surgical indication due to size. She was usually medicated with dabigatran 110 mg twice daily (bid.), bisoprolol 2.5 mg once daily (od), enalapril plus lercanipine 10/10 mg od, atorvastatin 20 mg od, and fluoxetine 20 mg od. Two days before the beginning of symptoms, dabigatran was changed to enoxaparin for short-time ovarian surgery. Although prescribed the adjusted dose for her weight (100 mg od), the patients started, by lapse, treatment with twice the prescribed dose (200 mg od) of subcutaneous enoxaparin. She had no history of trauma or drinking.

On initial evaluation, the patient presented with a Glasgow coma scale score of 15, pain (level 7-8 on visual analogue scale for pain) and hypertension (blood pressure 164/73 mmHg). There were no abnormal findings on neurological exam. She was medicated with 8 mg of intravenous lornoxicam and 5 mg of oral diazepam. The complete blood count showed normal hemoglobin (13.6 g/dL) and mild thrombocytopenia (118x10^9/L) and no alterations on the coagulation screen and biochemistry. The cervical radiography excluded fractures and showed suggestive changes of arthroses and the cranial computed tomography (CT) was normal.

During the stay in the ED, the patient developed decreased muscle strength in all limbs and global hypoesthesia, whereby it was requested a cranial CT.

The patient became drowsy with a sustained neurological decline which culminates in quadriplegia, hypoesthesia below D5 level and absent deep tendon reflex. She was medicated with a bolus of 200 mg of hydrocortisone. Cervical CT described posterior interapophysary arthrosis between C3-C4 conditioning stenosis with no other alterations, and lumbar puncture was unremarkable.

The patient presented a clinical improvement in mental status (after the administration of hydrocortisone) with recovery of muscular strength with a grade 4/5 in Medical Research Council scale in the upper limbs and a grade 3/5 in the lower limbs, although not sustained, with some fluctuations during the stay in the ED. Despite the improvement of the clinical status, she developed spontaneous muscular myoclonus mainly on the left hemibody and urinary retention.

A MRI of the cervical and thoracic spine was then performed and revealed the presence of a hyperintense collection on T1 and T2 weight images from C2 to D3 level, suggestive of acute epidural hematoma and with signs of mass effect (Fig. 1). Given the diagnosis, the patient was immediately transferred to a neurosurgery centre.

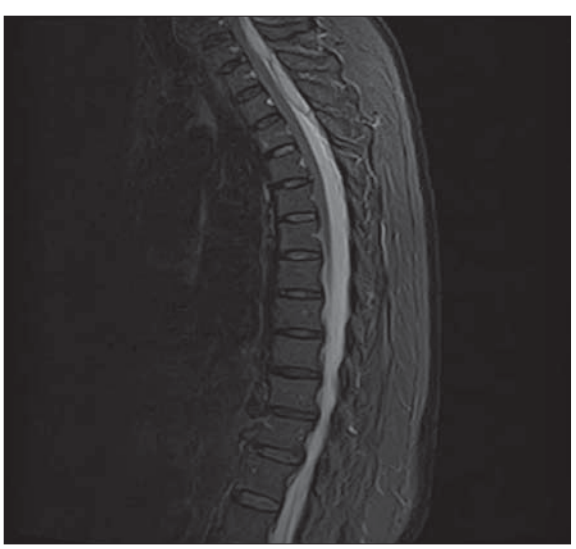

Figure 1. Sagittal short tau inversion recovery magnetic resonance imaging of the cervical spinal cord showing an acute cervical epidural hematoma.

During the following seven days, the patient remained with conservative treatment and under surveillance in the neurosurgery ward. She presented a progressive clinical improvement being able to walk with assistance and

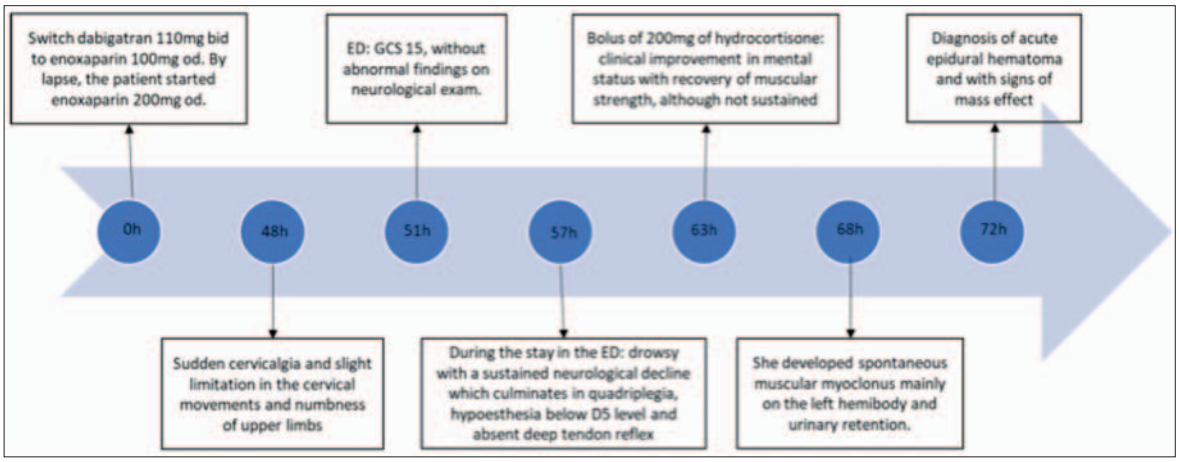

Figure 2. Temporal development of the symptomatology due to acute epidural hematoma.
Bid: twice daily; od: once daily; ED: Emergency Department; GCS: Glasgow Coma Scale.

showing a regression size of the epidural hematoma on the control MRI.

The patient complied with an intensive functional rehabilitation plan, maintaining progressive improvement. She was discharged four weeks after admission without neurological deficits.

Before restarting anticoagulation treatment, she performed a MRI which showed the complete absorption of the haematoma, an echocardiogram which revealed mild dilatation of the left atrium and normal left ventricular global systolic function and a 24-hour Holter monitoring which recorded continuous atrial fibrillation rhythm. With a CHADs2VASC score of 3 (arterial hypertension, age and female gender) and a HAS-BLED score of 1 (prior bleeding), the patient restarted anticoagulation treatment with dabigatran 110 mg bid and propafenone 150 mg bid, as a rhythm control strategy. After one year of follow up, the patient is in sinus rhythm and without haemorrhagic complications.

Discussion

A spine epidural hematoma is a rare (1 per million estimate incidence[4]) and significant neurological disorder. The association with anticoagulant therapy is extremely rare.[5] Even though the aetiology remains unclear, there are some hypothesis to explain it. According to Bruyn and Bosma[6] increased intrathoracic and intra-abdominal pressure (like straining, bending, coitus, coughing or sneezing) leads to an increase in intravenous pressure in valveless and thin-walled epidural veins leading to rupture. However, in the cervical region, the rapidity of development of epidural hematoma points to artery origin. According to Beatty and Winston,[7] free anastomotic arteries which run in the epidural space and connect with radicular arteries are responsible for arterial bleeding in cervical epidural hematomas. SSEH could occur in any level of the spinal cord, although it seems to appear more often in the posterior cervicothoracic (C5-T2) and thoracolumbar (T10-12) levels, due to the presence of Hoffmann ligaments connecting the ventral dura to the posterior longitudinal ligament.[8]

There are some factors which have been associated to spontaneous hematoma, as anticoagulant therapy for prosthetic cardiac valves, therapeutic thrombolysis for acute infarction, long-term aspirin use, haemophilia B, factor XI deficiency and vascular malformations.[3] In our case, the patient was on anticoagulant therapy, and she had recently changed from a direct-acting oral anticoagulant to a low molecular weight heparin and in supratherapeutic dose by fault. This may have contributed to this event.

SSEH symptoms will depend on the site and severity of spinal cord compression, ranging from simple back pain to acute quadriplegia. Initially, what appeared to be a simple cervicalgia, presented an unusual evolution, with a floating clinic. The risk of unusual manifestations is that they can delay or lead to a misdiagnosis. It is described that SSEH can mimic situations such as acutely ruptured cervical disc, epidural neoplasia, transverse myelitis, dissecting aortic aneurysm, congenital cysts, spondylitis, or epidural abcess.[9] As mentioned above, MRI is the best method to establish the diagnosis. CT was performed before the MRI since it was the only imaging method available at the time of admission, which was useless to the diagnosis, highlighting the role of MRI for evaluation

of spinal cord diseases. This delay in diagnosis was the reason why protamine sulphate was not given since the half-life of enoxaparin is 12 hours.

Usually, in the first twenty-four hours, an epidural hematoma appears as an hyperintense area on T2-weighted images in combination with focal areas of hypointensity and isointensity on T1-weighted images. After a few days, the hematoma appears as an hyperintensity area on T1-weighted images and as hypointensity area on T2-weighted images because of the presence of intracellular methemoglobin.[10] This evolution over time, could explain why the MRI performed to our patient, revealed a hyperintense collection on T1 and T2-weight images, once it was performed several hours after patient admission.

After a long period time that routine use of steroids was abandoned due to a higher incidence of complications and no evidence of efficacy,[11] recently cross-sectional study[12] and a Cochrane Database of Systemic Reviews article,[13] described significant neurological improvement when the patients were treated with high doses of methylprednisolone within eight hours of injury. In our case we have seen a clinical improvement after the steroid bolus, not being able to conclude whether there was a causal relationship (perhaps reducing oedema) or whether it was a mere co-occurrence.

Surgical procedures as decompressive laminectomy and hematoma evacuation are the standard treatment for SSEH (Table 1). Incomplete deficits and an earlier intervention are associated with better outcomes.[2] Even so, there is an increasing number of cases reports which were managed conservatively.[14] Rapid improvement of neurological deficits, coagulopathy, and the refusal of surgery are some scenarios which may support the conservative treatment.[15] In our case, the rapid recovery and the time between the onset of symptoms and the arrival to the neurosurgical centre could explain the choice of treatment.

The possibility to re-initiate anticoagulant therapy (and the time frame to do it) in patients with central nervous system haemorrhage is often a dilemma. We waited for the complete resolution of the hematoma, and chose dabigatran as the anticoagulant drug, since it is the only direct-acting oral anticoagulant with an available antidote.

Conclusion

This case describes a cervical epidural hematoma associated with high anti-coagulant doses in a patient with no other signs of haemorrhagic complications. Prompt suspicion of spinal cord injury is mandatory in patients with a rapid clinical condition of typical symptoms. MRI is of unquestionable added value and necessary for the diagnosis. Prompt evaluation in order to consider surgical management is of high importance. ■

Responsabilidades Éticas

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Table 1. Clinical cases described on literature.

Paper	Sex	Age	Location	Medication	Treatment
Akpınar A, Celik B, Canbek I, Karavelioglu E[16]	Male	80	T1 – T12	-	Surgery
Sarwal G, Dandurand C, Lee A, *et al*[17]	Male	81	T1 – T12	Rivaroxaban plus Aspirin	Surgery
Mathais Q, Esnault P, Cruc M, *et al*[18]	Male	78	C4 – T5	Dabigatran	Surgery
Girithari G, Coelho dos Santos, I, Alves T, Claro E, Kirzner M, Massano A[19]	Woman	57	T4 – T8	Warfarin	Surgery

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