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Spontaneous cervical epidural hematoma with incomplete Brown-Sequard Syndrome

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Introduction

Cervical epidural hematoma, without a traumatic etiology, is a rare clinical entity. In the neurosurgical literature it is called spontaneous cervical epidural hematoma (SCEH) and, both the venous and arterial theories provide plausible explanations for its occurrence. The potential causes include coagulopathies, hypertension, vascular malformations and conditions that increase venous pressure such as sneezing, coughing or vomiting (25).

The clinical presentation is usually characterized by acute radicular pain followed by progressive paralysis and sensory loss of symmetric nature. The Brown-Sequard syndrome is an exceptional result of SCEH (7, 16, 28). Surgical evacuation of the hematoma is the standard therapy, although conservatively treated SCEH cases have been reported (9). We presented a case of a cervical spontaneous epidural hematoma presenting with an incomplete Brown-Sequard syndrome and treated by surgical evacuation.

Case report

The patient, a 68-year old female, presented to our hospital with a history of acute neck pain and left side weakness, symptoms that started 3 weeks earlier, after she did a mild physical effort. She complained pain on her posterior neck and left shoulder while neck rotation. Her past medical history was significant for hypertension, she had taken aspirin due to chronic cardiac ischemia.

On neurologic examination, she had 3/5 motor strength in muscle groups of the left upper and lower limbs and reduced sensation of pain contralaterally below the C5 level (incomplete Brown-Sequard syndrome). Sensation of light touch was present. Deep tendon reflexes were brisk and plantar response was extensor on the left side. She had normal coagulation values. At her first admission, in another hospital, the patient, known with a prolonged history of hypertension, was misdiagnosed as infarction on right hemisphere and was delayed in diagnosis.

A computed tomography (CT) scan of the brain was normal. Cervical magnetic resonance imaging (MRI) showed a large left posterolateral extradural mass extending from C2 to C6 with spinal cord compression and rightward displacement. The mass demonstrated low signal intensities on T1-weighted images and intermediate to high signal intensities on T2-weighted images (Figures 1A, 2A and 3A).

No contrast enhancement or signs or signs of vascular malformation were found. A cervical spinal angiography was performed in order to diagnose an underlying pathology but it was normal (Figure)

The patient underwent a C3-C5 hemilaminectomy of left side. A dark blood clot was noted in the epidural space and was evacuated under microscope. No evidence of vascular malformation was found.

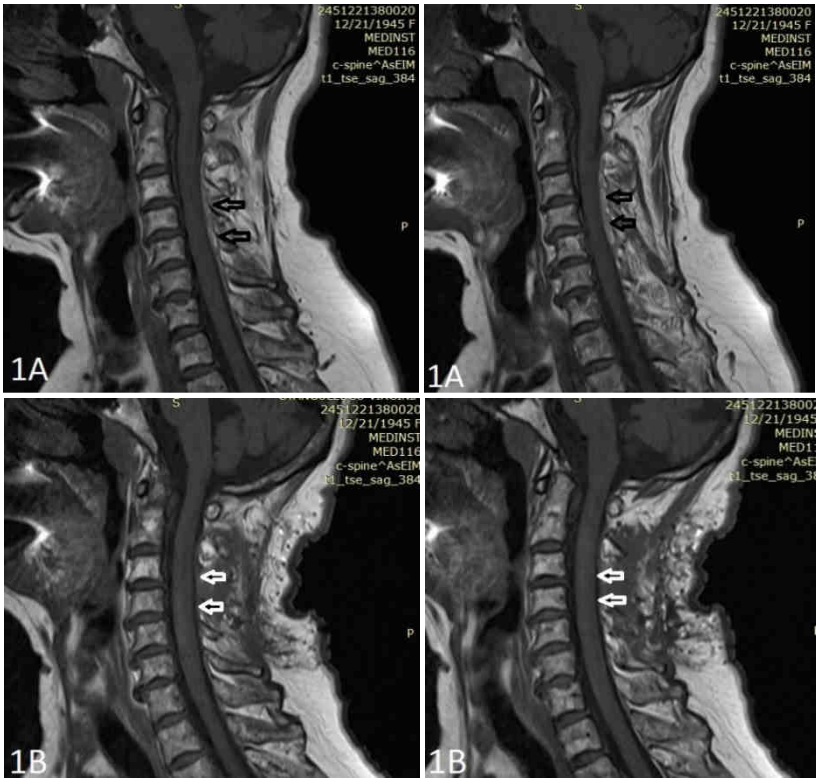

Figure 1 (A) - Preoperative T1 weighted saggital images (black arrows) show intermediate signal left extradural mass extending from C2-C6 (higher intensity that the signal of the spinal cord) (B) - Postoperative T1 weighted saggital images after evacuation of hematoma (white arrows)

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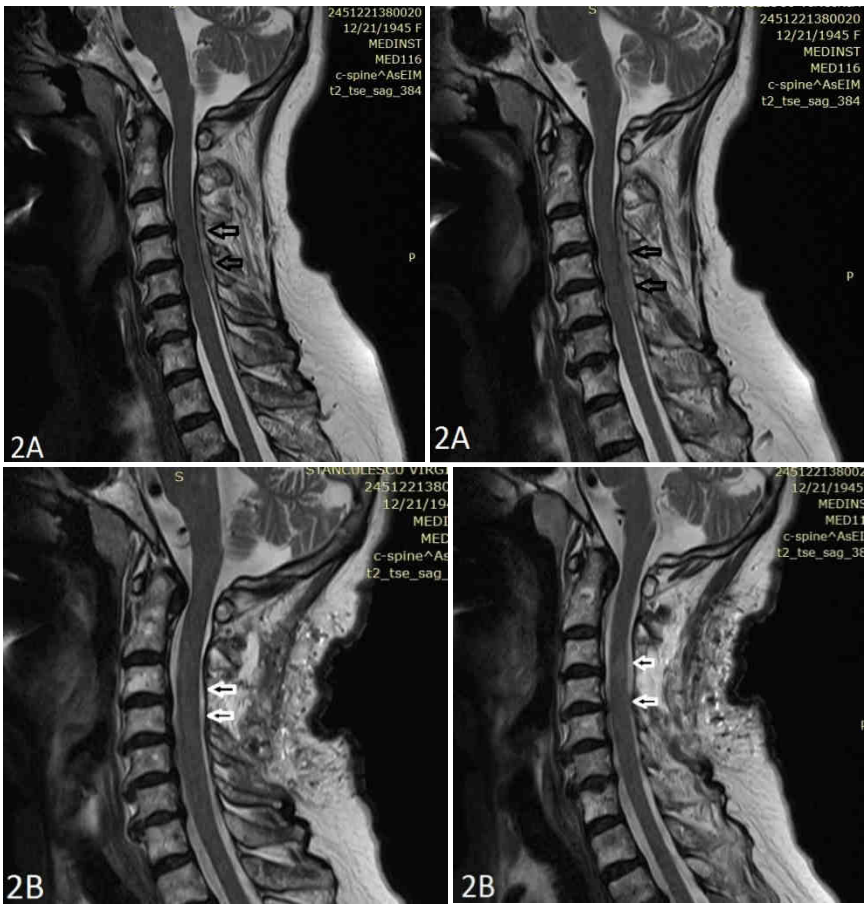
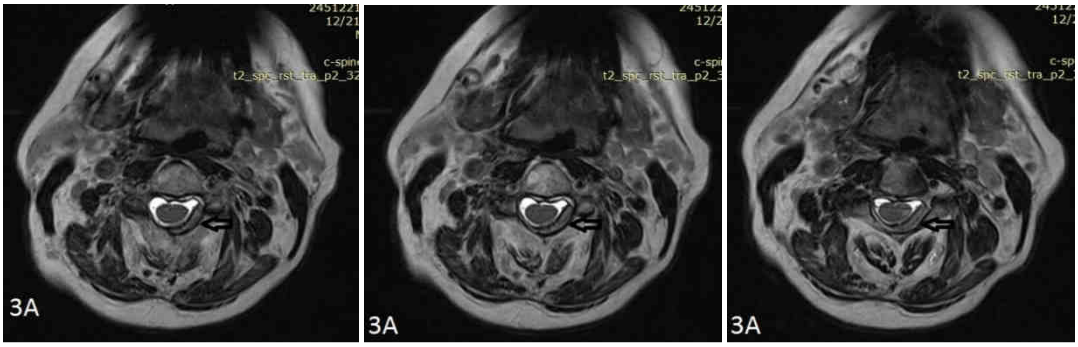


Figure 2 (A) - Preoperative T2 weighted saggital image show intermediate signal of an extradural mass extending from C2-C6 (black arrows); posterior compression of the spinal cord with interruption of the intense signal of the CSF at this level. (B) - Postoperative T2 weighted saggital images after evacuation of hematoma and decompression of cervical spinal cord; presence of the intense signal of CSF in the posterior aspects of the C2-C6 vertebrae (white arrows)



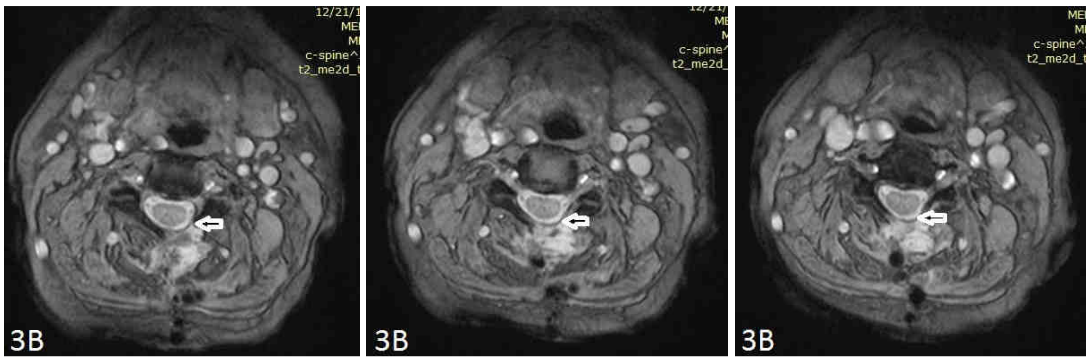


Figure 3 (A) - Preoperative T2 weighted axial images show intermediate signal of an left posterolateral extradural mass with spinal cord compression (black arrow), (B) - Postoperative T2 weighted axial images showing no compression on cervical spinal cord and resumption of CSF flow after evacuation of the hematoma (white arrow)

Postoperatively, the patient recovered well until prompt return of her sensory symptoms. Her motor function gradually improved within the next 48 hours. Postoperative MRI showed complete evacuation of the hematoma (Figures 1B, 2B and 3B). On her six months follow-up, she was doing well, with no residual neurological deficits.

Discussion

Spontaneous cervical epidural hematomas (SCEF) are well recognized but rare surgical conditions (28). It was first described in 1869 by Jackson (19) and the first surgical approach was performed in 1946 by ver Brugghen (31). One of the first extensive reviews on spontaneous spinal epidural hematoma was reported by Groen in 1996 (15). A more recent one, by Kim et al in 2011 (9), identified a total of 107 cases of spontaneous spinal epidural hematoma published in neurosurgical series between 1996 and 2011.

In the majority of reports, the hematoma is considered to be "spontaneous" if it develops without known case (i.e. trauma, fractures, spinal lesion or medical procedure). Usually, SCEH develops because of a predisposing condition, including anticoagulant therapy (11, 24), vascular malformations (5,30), neoplasm (17), immunovasculitis (10,20), hypertension (11),or bleeding tendencies of all kinds (12,26). However, the cause of bleeding remains unknown in 40% of the cases and are called idiopathic spontaneous cervical epidural hematoma (11). In our case, the patent had a medical history for hypertension..

It is still debated if the bleeding that lead to the occurrence of spontaneous epidural hematoma is arterial or venous. Both the venous and arterial theories provide plausible explanations, but, most likely, the SCEH results from a venous bleeding, because the epidural veins have thin walls and no valves (32). The posterior epidural venous plexus (22) lies directly between the dura and the ligamentum flavum making rupture more likely than in the anterior venous plexus which is covered by the posterior longitudinal ligament. Given that the epidural venous plexus is devoid of valves, sudden changes of

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intra-thoracic and intra-abdominal pressure, after Valsalva maneuvers can determine lacerations in the venous plexus, causing epidural hemorrhage.

In contrast, Beatty suggested that bleeding occurs from epidural arteries (6). He argued that hemorrhage from an epidural vein would not create enough pressure to compress the dural sac, observing that the normally expanded epidural sac may tamponade epidural bleeding, encountered during surgical procedures (6). Beatty and Winston sustained that is more logical that epidural arteries are likely causative, especially in the light of the compression of the cord that can occur and the posterolateral location of most cervical epidural hematomas (2). They have explained that the C6 and C7 segments are the most common region of cervical epidural hematomas and extreme compelling movements could therefore cause tearing of these arteries (2). Lowery has defined an active bleeding arterial structure under the hematoma in one of the operated cases (21).

The clinical picture of SCEH is characterized by sudden onset of neck pain, with or without radicular radiation, followed by rapidly progressive symptoms and signs of cord compression. Rarely, however, patients may present with slowly progressive symptoms which can lead to difficulties in diagnostic (3). Pain radiation varies according to the localization of the hematoma on the spinal cord and nerve roots. The second most common symptom is weakness of the limbs, seen below the compressed spinal cord. The neurologic deficits may be mild or complete (11,24) and, based on the location of the hematoma, they can be bilateral, show features of a Brown-Sequard syndrome (7,30), or, rarely, anterior cord syndrome (11). Paresis increase within minutes or days, or, rarely, recover spontaneously (2). We presented the case of a female patient, with the neurological signs of an incomplete Brown-Sequard syndrome. Cervical epidural hematoma presenting with Brown-Sequard syndrome is a rare condition (8), but has been reported in the literature (16, 27, 28, 32). This unusual presentation can be confused with a cerebrovascular accident, as in our case, when the patient, known with a prolonged history of hypertension, was misdiagnosed as infarction on right hemisphere and was delayed in diagnosis. Acute cervical disc herniation, transverse myelitis, spondylitis, epidural neoplasia, dissection of aortic aneurysm should be considered in the differential diagnosis (1, 28).

Magnetic resonance imaging (MRI) is considered the first choice of diagnostic method for SCEH. It gives accurate information not only concerning the location and extension of the hematoma, but also the degree of the cord compression, as well as any preexisting lesion that might have been the source of the bleeding, such as arteriovenous malformations (18, 28). Before 48 hours, the signal intensity of the acute cervical epidural hematoma on T1-weighted images is varying from hyper-intense to iso-intense. On T2-weighted images, acute cervical epidural hematoma demonstrates focal hypo-intensity, in contrast with the heterogenous hyper-intensity of the cord. Old hematoma (more than 21 days) shows, in contrast, high signal

intensity on the T2W1 and mixed (low and iso-) signal intensity on T1W1 (18). On T1-weighted post-contrast images, peripheral contrast enhancement due to adjacent dural hyperemia may be seen (4, 28). The T1 and T2 signals of SCEH vary based on the clot, age, size and oxygenation (14). MRI has greatly facilitated the diagnostic of SCEH allowing for more cases, with a benign natural course to be diagnosed (20).

Surgery is the treatment of choice in SCEH. Emergent surgical decompression and evacuation of the hematoma is needed, especially in cases with neurological deterioration. Some authors consider that total laminectomy is the best choice as a surgical approach but hemilaminectomy on two or third cervical levels, can be preferred according to the localization of the hematoma (1, 28, 32). In our case, we performed a C3-C5 left hemilaminectomy, and under microscope magnification we completely evacuate the left posterolateral epidural hematoma.

In recent years, there have been increased reports of spontaneously resolving spinal epidural hematomas, due to quick diagnosis by higher resolution MRI (9). A conservative treatment under close neurologic observation is recommended for patients with no, or mild neurologic deficits and for patients who show definite progressive improvement prior to the MRI diagnosis of SCEH (9, 13, 27).

The major factors determining neurological recovery after spontaneous cervical epidural hematoma are the localization of the hematoma (cervical segments involved), the preoperative neurological condition and the operative interval (9, 15). Initial neurological dysfunction was the strongest predictor for a patient's outcome. Patients presenting with severe or worsening conditions usually did not recover as wells as those presenting with minor symptoms (9). Furthermore, the timing of decompressive laminectomy and evacuation of the hematoma was more significant factor in patients initially presenting with Frankel grades A or B. Patients with a better neurological function (scores of D or E) frequently showed substantial improvement regardless of time of operation (9, 28, 32). In complete preoperative sensori-motor loss, surgery within 36 hours correlated with favorable outcome; in incomplete preoperative sensori-motor deficit, favorable outcome correlated with surgery within 48 hours (15). Also, those patients with a shorter interval to operation had a better prognostic than those with longer intervals. Cho Y.E et al (29) and Kim DK et al (9), on a large reviews of cases with spontaneous spinal epidural hematomas, showed that neurological outcome was good in those patients that had their hematoma evacuated within 24 hours, and the patients with a preoperative incomplete neurological deficits, who had a surgical operation performed within 12 hours, had an excellent surgical outcome. In our case, surgery was performed within 36 hours after admission, but eight days after the development of hemiparesis. Postoperative, a gradual recovery in neurological status and improvement of sensation and motricity was observed within the next 48 hours.

Even patients with significant comorbidities did achieve functional recovery,

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comparable with patients without comorbidities. A relevant past medical history does not predict poor outcome, but may be associated with worse initial neurological dysfunction than others (9, 20).

Conclusions

Spontaneous cervical epidural hematoma is a rare neurosurgical emergency and prompt diagnosis using cervical MRI is very important. Both the venous and arterial theories provide plausible explanation for the occurrence of SCEH. The development of a Brown-Sequard syndrome due to a SCEH is very are, and is usually incomplete. The clinical presentation of the cervical epidural hematomas with hemiparesis can be misdiagnosed as a cerebrovascular accident. Surgical decompression must be preferred in patients presenting with neurological deterioration. Patients with less initial neurological deficits and those with shorter time to operation show better neurological prognostic. In our opinion, a prompt diagnostic and an appropriate surgical treatment is essential for a favorable functional neurological recovery.

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