

## Case Report

# Spontaneous Bilateral Supratentorial Subdural and Retroclival Extradural Hematomas in Association with Cervical Epidural Venous Engorgement

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We describe a case of 36-year-old man who presented with a subacute headache preceded by a 1-month history of posterior neck pain without trauma history. Head and neck magnetic resonance imaging (MRI) studies disclosed bilateral supratentorial subdural and retroclival extradural hematomas associated with marked cervical epidural venous engorgement. Cerebral and spinal angiography disclosed no abnormalities except dilated cervical epidural veins. We performed serial follow-up MRI studied to monitor his condition. Patient's symptoms improved gradually. Serial radiologic studies revealed gradual resolution of pathologic findings. A 3-month follow-up MRI study of the brain and cervical spine revealed complete resolution of the retroclival extradural hematoma, disappearance of the cervical epidural venous engorgement, and partial resolution of the bilateral supratentorial subdural hematoma. Complete resolution of the bilateral supratentorial subdural hematoma was confirmed on a 5-month follow-up brain MRI. The diagnosis and possible mechanisms of this rare association are discussed.

**KEY WORDS :** Supratentorial subdural hematoma · Retroclival extradural hematoma · Cervical epidural venous engorgement

## INTRODUCTION

Intracranial subdural hematoma generally occurs following head trauma<sup>10,17,24,25</sup>. Spontaneous intracranial subdural hematoma has been infrequently reported in association with coagulopathy, ruptured cerebral aneurysm or arteriovenous malformation<sup>3,12,13,16,19,20</sup>. Spontaneous subdural hematomas of arterial origin are also rarely reported<sup>15,27,28</sup>. Extradural hematoma of the retroclivus is extremely rare and is usually found in severely injured patients<sup>6,9,15</sup>. Enlargement of cervical epidural vein are rarely encountered on imaging studies. It may enlarge in pathologic conditions such as a vascular malformation or a jugular venous obstructive lesion<sup>4,8</sup>.

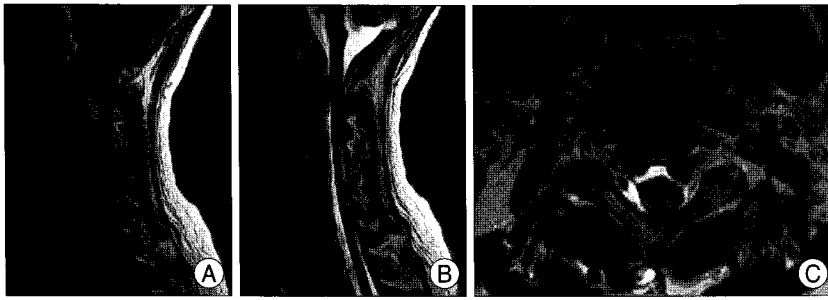
In this report, we present an exceptional case of the patient with spontaneous bilateral supratentorial subdural

and retroclival extradural hematomas accompanied by cervical epidural venous engorgement. We discuss the diagnosis and possible mechanisms of this rare association.

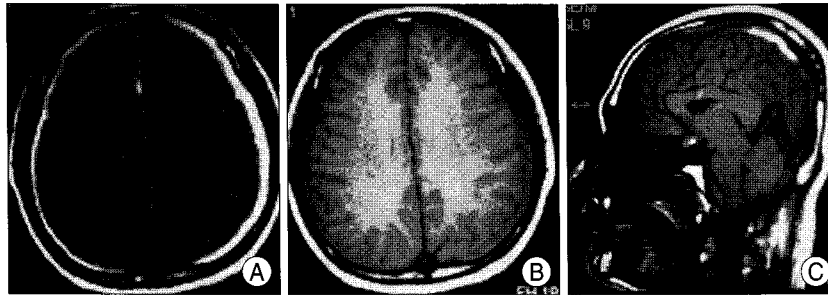
## CASE REPORT

A 36-year-old man was admitted with a 5-week history of posterior neck pain and a 1-week history of headache. Five weeks before admission the patient experienced acute onset of severe posterior neck pain. He had presented to the local clinic. MRI of the cervical spine was performed, which showed dilated cervical epidural venous plexus (Fig. 1). He was treated with analgesics but his symptom persisted. Four weeks after his initial presentation, the patient had a gradual onset of progressive headache. The patient was evaluated at another hospital for headache. MRI of the brain revealed bilateral supratentorial subdural and retroclival extradural hematomas (Fig. 2). The patient was then transferred and admitted to our institution. The patient had no significant medical history and denied any preceding trauma. Neurological examination was normal. Routine laboratory studies were normal. Cerebral and spinal angiography disclosed dilated cervical epidural veins at the level of atlas and axis.

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**Fig. 1.** Cervical MRI taken at local clinic. A and B : Sagittal T1 and T2-weighted MRI show mass of mixed signal that are located at anterior cervical epidural space. C : Axial T2-weighted MRI shows bilateral dilated epidural cervical venous plexus.



**Fig. 2.** Local clinic brain MR images. A and B : Axial T2 and T1 weighted MRI show subdural hemorrhage (SDH) of high and low signal that are located at supratentorial area. C : Sagittal T1-weighted MRI shows extradural hematoma that are located at retroclival area.



**Fig. 3.** Cerebral and spinal angiography show dilated cervical epidural veins at the level of atlas and axis. The internal jugular veins were normal, but asymmetrical. No intracranial or spinal arteriovenous fistula was identified.

The internal jugular veins were normal, but asymmetrical (Fig. 3). No intracranial or spinal arteriovenous fistula was identified. The patient was observed closely with gradual symptomatic improvement. Follow-up MRI of the brain and cervical spine obtained 9 days after admission showed marked regression of cervical epidural venous engorgement (Fig. 4). Bilateral supratentorial subdural and retroclival extradural hematomas remained unchanged in size (Fig. 5). He was discharged from the hospital three weeks after admission in a stable condition. Serial repeat radiological studies were performed after his discharge. A 3-month follow-up MRI of the brain showed complete resolution of the retroclival extradural hematoma (Fig. 6). Total absorption of the supratentorial subdural hematomas was confirmed

on a 5-month follow-up brain MRI (Fig. 7).

## DISCUSSION

The cervical epidural venous system provides independent accessory venous drainage from the intracranial compartment. There is no posterior epidural vein in the cervical spine as in the lumbar spine. The entire cervical epidural vein system is located in the anterolateral part of the spinal canal. Enlarged cervical epidural veins are usually caused by an underlying vascular lesion such as a malformation or a jugular venous obstructive lesion<sup>4,8</sup>. Dickman et al.<sup>8</sup> described a case of myelopathy developing from epidural varicose veins compressing the cord at the cervicothoracic junction and improving after surgery. However, it can also be caused by

intracranial hypotension. The spinal system may function as an alternative cranial drainage system under some circumstances and the cervical venous system can affect intracranial pressure<sup>1</sup>. Only two reports exist in the literature of engorgement of the cervical epidural venous plexus associated with intracranial hypotension<sup>5,26</sup>. Clarot et al.<sup>5</sup> reported giant cervical epidural veins after lumbar puncture in a case of intracranial hypotension. Shinaver et al.<sup>26</sup> reported that a case of engorged anterior epidural venous plexus mimics an anterior epidural mass that was thought to represent an epidural abscess based on preoperative MRI finding. Therefore, epidural venous engorgement should be suspected in the clinical finding of intracranial hypotension.

Retroclival extradural hematoma is extremely rare and is usually found in severely injured patients<sup>15</sup>. The diagnosis of retroclival extradural hematoma can be easily confirmed by MRI of the brain. Sagittal sections are crucial for correct diagnosis. In our search of the literature, only eight cases have been reported<sup>14</sup>. All of the cases previously published had many features in common. All cases were children who were severely injured and sustained a major injury to the craniocervical junction. However, our presented case was did not have a history of injury and had occurred spontaneously. The mechanism of retroclival extradural hematoma may be thought to be caused by dissection of adjacent dura over the clivus due to markedly engorgement of cervical epidural veins associated with spontaneous intracranial

hypotension. Until now, there has been no report of retroclival hematoma due to spontaneous intracranial hypotension.

Subdural hematoma is commonly regarded as a complication of a head injury and in case of non-trauma, subdural hematoma is associated with the rupture of cerebral aneurysm or vascular malformation. It may also be caused by intracranial hypotension due to a shunt operation for hydrocephalus and cerebrospinal fluid leakage after head trauma or lumbar puncture. A review of the literature has revealed a few case reports of spontaneous intracranial hypotension (SIH) associated with subdural hematoma<sup>7,18</sup>.

SIH is a syndrome of low cerebrospinal fluid (CSF) pressure (below 70 mmH<sub>2</sub>O) characterized by postural headaches in patents without any history of dural puncture, surgery or penetrating trauma<sup>21,23</sup>. The pathogenesis is usually considered to be an occult CSF leak through small defects in the meninges with a resultant decrease in CSF volume and pressure<sup>11,22</sup>.

We consider the causative factor of cervical epidural venous engorgement, spontaneous subdural hematoma and retroclival extradural hematoma as SIH based on the evidences that we described above.

There are several reasons why the causative factors are considered as SIH in the current case. Firstly, all three types of clinical symptoms can be caused by SIH. Secondly, most of the cases of SIH have a benign course and they are improved by the conservative care<sup>3</sup>. Also, in the current case, the clinical findings and symptoms are improved by the conservative care.

Through a history taking, neurological examination and several radiologic findings, no common etiologies of them have been found. To put this in another way, the possibility for common etiologies could be ruled out. Fourthly, three abnormal symptoms are manifested at the same time. Lesions were present in such an extensive areas as supraten-

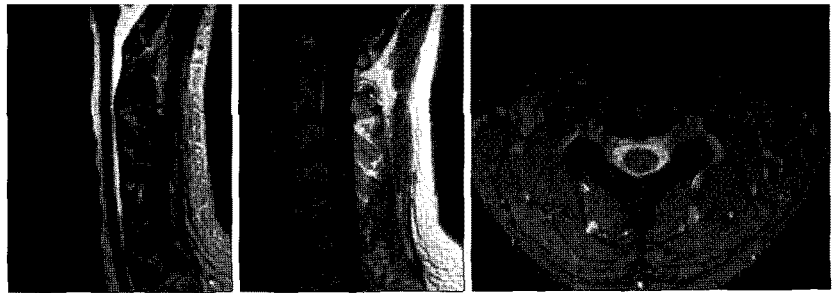


Fig. 4. Follow-up MRI of cervical spine obtained 9 days after conservative treatment shows marked regression of cervical epidural venous engorgement compared with previous film.

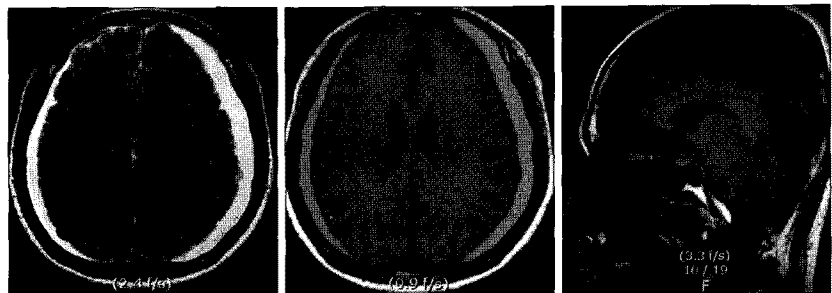


Fig. 5. Follow-up MRI of brain obtained 9 days after conservative treatment shows that bilateral supratentorial subdural and retroclival extradural hematomas remained unchanged in size compared with previous film.

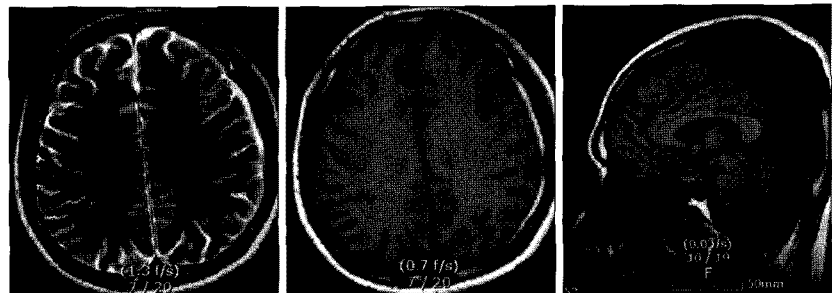


Fig. 6. A 3-month follow-up MRI of brain shows complete resolution of the retroclival extradural hematoma.



Fig. 7. A 5-month follow-up MRI of brain shows complete resolution of the supratentorial subdural hematoma.

torial, infratentorial and cervical spine, not a focal region. These findings are suggestive of SIH. Fifthly, the clinical findings were improved in a sequential manner. These findings also indicate that a single etiology was involved. In other words, while the cervical epidural vein engorgement was improved, the retroclival extradural hematoma was also

improved and this was followed by the improvement of bilateral supratentorial SDH. Based on these findings, we considered the current case to be due to SIH.

To the best of our knowledge, the present case represents the first recognized case of a spontaneous bilateral supratentorial subdural and retroclival extradural hematomas accompanied by cervical epidural venous engorgement due to SIH at the same time.

In the current case, the clinical symptoms were improved by the conservative treatment. We conclude that conservative treatment and close observation are more beneficial than aggressive treatment for the current case. Although rare, however, a more detailed examination may be mandatory to identify CSF leaking point in recurred cases.

## CONCLUSION

We report an extremely rare case of spontaneous bilateral supratentorial subdural and retroclival extradural hematomas in association with cervical epidural venous engorgement at the same time. It is important to consider the possibility of SIH as the causative factor of these clinical finding.

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