Case study

Neurologic Complications After Repeat Epidural Blood Patch in Recurrent Spontaneous Intracranial Hypotension : A Case Report

Abstract

A 61-year-old male patient presented to Guro Korea University Medical Center with a one-month history of increasingly severe headaches that were aggravated by an upright position and alleviated by a supine position. The patient had no other specific underlying disease. Epidural blood patch was repeatedly performed several times because headache was not fully subsided, 10 minutes after fourth EBP, the patient showed a decrease in motor grade 1 in both lower limbs, Lt. Upper limbs and Grade 3 for Rt. upper limbs. Urgent surgical evacuation of spinal epidural hematoma was conducted. After operation, patient recovered from neurologic deficit.

While epidural blood patches are generally considered safe and effective for treating SIH, like any medical procedure, there can be potential complications. It's important to note that complications are relatively rare, and the benefits of the procedure often outweigh the risks. When we repeatedly perform epidural blood patches, more caution is required. Careful evaluation of absolute indications is necessary to prevent additional complications and possible complications should be seriously considered.

Keywords

Epidural blood patch, quadriplegia, neurologic deficit, spontaneous intracranial hypotension, CSF leakage

Introduction

Spontaneous intracranial hypotension (SIH) occurs due to a cerebrospinal fluid (CSF) leak in the spine, leading to CSF hypovolemia and subsequent intracranial hypotension[1]. This

condition results from the loss of CSF volume that normally surrounds and cushions the brain and spinal cord.

Epidural blood patch (EBP) is an effective treatment for spontaneous CSF leak syndrome patients which is characterized by occipital or frontal headaches that improve when lying down and worsen when in an upright position [2].Usually, about 20 mL of autologous blood that is drawn and injected into the epidural space forms a clot that can seal the leak and help restore normal CSF pressure[3, 4].The normal range for CSF is reported differently in various sources, with most reporting a normal range of 7-18 cmH2O in adults[5]. While an EBP is considered effective treatment for CSF leakage, unexpected complications may occur. Common complications may include headaches, neck pain, and back pain[6]. Although rare, serious complications such as chronic adhesive arachnoiditis, subdural or spinal hematoma, seizures, infections, and intracerebral hemorrhage can also occur[7]. In most cases, these complications can be managed through conservative treatments, and in the instance of severe complications, prompt intervention is often instrumental in preventing the development of significant issues.

Case presentation

A 61-year-old male patient presented to Guro Korea University Medical Center with a one-month history of worsening severe headaches that were aggravated by an upright position and alleviated by a supine position. The patient had no other specific underlying disease and recent trauma history.

Diagnostic workup included brain Magnetic Resonance Imaging (MRI). Brain MRI revealed diffuse subdural hematoma (SDH) and dural enhancements along both the cerebral convexity, falx and tentorium with suspicious dilatation of venous sinuses and sagging of the midbrain. A Computated Tomography (CT) myelography was performed to differentiate CSF leakage, revealing an extensive CSF leakage extending from C2/3 to T7/8. Based on diagnostic imaging, an EBP was performed at the T1/2 level. The patient's headache gradually improved. The patient was prescribed naproxen and discharged.

Nine days after the first procedure, the patient visited the outpatient clinic and expressed a recurrence of headache. The headache was sharp, stabbing with a Numeric Rating Scale (NRS) of 4 and exacerbated by movement. The follow-up CT showed increased in extent of

diffuse chronic stage of SDHin both cerebral convexities. Consequently, we conducted a second EBP at the T1/2 level, which was at the same location as the first EBP, two weeks after the first EBP. Following the second EBP, a marked alleviation of the headache was noted, and the patient was discharged.

One month after the second procedure, a follow-up brain CT exhibited increased densities of SDH at the bilateral cerebral convexities. As a result, a third EBP performed at the C3-4 level. If CSF leakage was not seen on CT myelography after the third procedure, burr hole operation was planned for SDH. However, after the third EBP, although the headache was improved, the findings on CT myelography still showed contrast leakage at ventral aspect of epidural space, C2/3 and C6/7 to mid-thoracic level (Figure 1). Therefore, we decided to postpone the Burr hole operation and proceed with a fourth EBP.

3minutes after fourth EBP (Figure 2), the patient showed a decrease in motor grade 1 in both lower limbs, 5 minutes later, decrease in motor grade 1 in Lt. upper limbs and grade 3 in Rt. Upper limb, and 7 minutes later, his speech began to slur. 10 minutes after EBP, the motor grade was grade 1 for both lower limbs and Lt. upper limbs and Grade 3 for Rt upper limbs. The patient's vital sign remained stable and the patient was alert. His pupils were isocoric, measuring 3 millimeters in diameter on both sides (3P/3P).

Spine MRI was taken immediately, and we found epidural hematoma with air bubbles from C2 to T4 posterior epidural space, compressing the spinal cord (Figure 3). Dexamethasone 10mg was administered to patient 1 hour after EBP for neuroprotection. Just before entering the operating room, the patient's motor function recovered to Grade 4, subjectively indicating an approximate 80% recovery. However, considering the lack of complete recovery of neurological deficit and significant cord compression, it was decided to proceed with the surgery in consultation with the neurosurgical team. An emergency operation was undertaken 2 hours after paralysis began. Urgent surgical evacuation of spinal epidural hematoma was conducted. Motor Evoked Potentials (MEP) and Somatosensory Evoked Potentials (SSEP) was monitored during the surgery. Hematoma was originated from externally injected autologous blood component, not hemorrhage from dura. Epidural hematoma was identified from C6 to T2. Hematoma was successfully evacuated without causing damage to the dura mater. Distal MEP was recovered after hemilaminectomy (C6-T2, Right) and hemipartial laminectomy (C5 Right, T3 Right). Proximal MEP showed signs of recovery after the evacuation of the epidural hematoma. CSF leakage from the cervical dura was suspected, but

the exact leakage site could not be located. After surgery, the patient's motor function showed full recovery as a Grade 5 in all four limbs and sensory function was also fully recovered. Two weeks after the surgery, the patient's headache completely improved, and a follow-up brain CT showed that the SDH was slightly resolved. The patient was discharged.

Discussion

SIH, also known as a spontaneous CSF leak syndrome, is a medical condition characterized by a low CSF pressure within the skull[1]. SIH usually occurs when there is a leakage of CSF from the spinal canal, reducing the pressure of CSF around the brain. This condition often causes a variety of symptoms that can vary in severity [2,8].SIH is a rare and challenging clinical condition both in terms of diagnosis and treatment [9].

Conservative management may include bed rest, increased fluid intake, and pain medications. In more severe cases, targeted interventions such as epidural blood patches, fibrin glue injections, or surgical repairs may be considered [3, 10].

Some SIH patients might have an underlying connective tissue disorder or other factors that predispose them to developing multiple leaks. However, EBP does not always provide a permanent solution, and may require repeated intervention [9].The primary treatment for refractory cases is the EBP, but its effectiveness varies [11].Repeated EBPs may be more frequently required in patients with SIH [12].

Prolonged INR and CSF leakage can be associated with repeated EBPs in patient with SIH [2]. However, in this case, no abnormal bleeding tendencies were detected in the blood coagulation test, indicating that repeated EBP were not associated with coagulation abnormalities.

Moreover, in some cases, as described in this report, complications may arise, including inadequate spread of blood components within the epidural space due to repeated EBPs[13]. If blood components injected during EBP do not disperse and accumulate, they can exert pressure on the spinal cord, causing a mass effect similar to an epidural hematoma, which may lead to neurological abnormalities. The coagulation function test results were within normal limits, suggesting that the epidural hematoma revealed by spine MRI was not attributed to the invasiveness. It is more likely that the patient's neurologic deficit is a result

of the autologous transplanted blood, which was confined to a localized area, rather than an epidural hematoma.

The inadequate spread of blood components within the epidural space may arise due to multiple reasons. Patient-specific anatomical variations, such as spinal curvature and epidural adhesions, could impede the uniform distribution of the injected blood[13]. Furthermore, the rate of injection might influence the ability of the blood to diffuse and cover the necessary area for effective sealing. It is also possible that repeated EBP procedures may have resulted in fibrosis or scarring within the epidural space, rendering it less amenable to blood spread[14]. Fibrosis-induced distortion of epidural anatomy can also contribute to EBP failure and subsequent complications [14].

In the case report by Iga K et al. [15], published in JA Clinical Reports, the authors present a noteworthy case involving a patient who underwent multiple EBP procedures due to unintentional dural puncture during combined spinal-epidural anesthesia for a scheduled cesarean section. After the repeated EBPs at the site of dural puncture, she reported neurological complications, such as radicular pains. The procedure led to neurological deficits due to issues related to blood component distribution and subsequent spinal cord compression. This case emphasizes the necessity for precise procedural considerations and vigilance when contemplating repeated EBP, especially in cases involving unintentional dural puncture. While our case and Iga K case both involve complications related to repeated EBP procedures, the underlying reason for EBPs was different. And in the Iga K case, conservative treatment like oral analgesic therapy was performed, and the neurologic symptoms were alleviated compared to the first time, so the patient declined further radiologic studies at the time and was discharged, which is different from our case where immediate emergency treatment was performed through surgical intervention.

The other case report by Carlswärd C presented the occurrence of chronic adhesive arachnoiditis following the administration of repeat EBPs [7]. A healthy, 29-year-old woman underwent two epidural blood patches due to severe post-dural puncture headache (PDPH)occuring after epidural analgesia for delivery. And she experienced lumbar pain and radiculopathy in both legs as well as pain radiating to the upper thoracic region. MRI showed changes with arachnoiditis and an intraduralhaematoma extending from T12 to S2. Conservative analgesic therapy with anti-inflammatory drugs, corticosteroids, and physiotherapy improved her condition slightly but during the following weeks her symptoms

worsened without significant relief. In the end, Follow-up MRI two years after the incident did not show further improvement and her physical condition remains unchanged.

The Carlswärd C case is an unfortunate case in which the patient's condition became chronic and the symptoms could not be relieved, affecting the patient's life. Compared to our case, both cases, Iga K and Carlswärd C, have something in common: complications that occurred after repeated epidural blood patches were performed.But, the reasons for performing the blood patch were different in the previous two cases, which were done due to PDPH following spinal and epidural anesthesia. However, in this case, the blood patch was performed due to the persistent SDH resulting from SIH.Furthermore, it is noteworthy to compare that in the previous two cases, headache improved after two sessions of EBP, while in this case, four EBPs were performed.

In our case, quadriplegia occurred due to compression of the spinal cord by the autologous blood components after four times EBPs. And the patient was recovered completely without sequelae after decompressive laminectomy. This is because, through prompt detection and response, surgery was decided and carried out immediately after rapid multidisciplinary consultation. Since this has not been reported before, we believe it is a good case that emphasizes the importance of close monitoring and rapid detection of patients after epidural blood patch, and the need for a multidisciplinary approach.

In cases where multiple EBP procedures are being considered, it is advisable to conduct thorough pre-procedural assessments, which may include additional imaging tests such as MRI [11]. MRI can help assess the patient's spinal anatomy and identify any structural changes or adhesions within the epidural space. It can reveal the presence of fibrosis or scarring that may affect the distribution of injected blood. By obtaining a clear understanding of the patient's spinal anatomy through imaging, healthcare providers can develop an individualized treatment plan. This plan can include selecting the most appropriate injection site, volume, and rate of blood injection to optimize the chances of a successful EBP. Choosing an appropriate injection site and technique can help optimize the spread of blood components. Additionally, consideration should be given to the volume and rate of injection, ensuring a controlled and uniform distribution of blood

While additional MRI can provide valuable insights into a patient's spinal anatomy and aid in the decision-making process for repeated EBP procedures, it's important to acknowledge

the potential cost limitations associated with this imaging modality. It should be reserved for cases where the benefits of the information obtained outweigh the associated costs and potential risks [16].

Successful outcome of this case can be attributed, in large part, to the vigilant neurological monitoring implemented post-EBP procedures. Continuous monitoring of the patient's neurological status allowed for the early detection of any emerging deficits, enabling rapid intervention before irreversible damage occurred. To identify subtle neurological changes and ensure comprehensive patient care, multidisciplinary approach involving anesthesiologists, neurologists, and neurosurgeons is necessary [17]. The decision to proceed with emergency surgery reflects the agility and expertise of the medical team involved. In cases where neurological deficits develop following interventions such as EBPs, prompt decision-making is crucial to mitigate potential complications. The surgical approach chosen likely depended on the patient's presentation, available imaging data, and the expertise of the surgical team. The case report should ideally elaborate on the factors considered when opting for surgical intervention.

In addition, we would like to earnestly emphasize, one of the most crucial aspects of managing patients undergoing multiple EBP procedures is thorough patient education and obtaining informed consent. Patients must be fully aware of the potential complications that can arise when EBPs are performed repeatedly. It is essential to explain that in severe cases or when complications occur, surgical intervention may be necessary to address spinal cord compression and neurological deficits.

Conclusion

This case is distinguished from previous reports in its unique context, involving a total of four EBPs performed due to recurrent SIH, resulting in quadriplegia due to spinal cord compression by autologous blood components. It is a rare but serious complication of EBP procedures in the management of recurrent SIH.

An understanding of the underlying anatomy, pathophysiology, and factors influencing EBP success is crucial for preventing adverse outcomes. When EBP is performed repeatedly, more efforts are needed to mitigate the risk of complications.Additionally, the importance of close monitoring after EBP should be explained to patients and the need for immediate reporting of

new or worsening symptoms should be emphasized.

Our experience highlights the necessity for close monitoring and swift detection of neurological changes following EBPs, emphasizing the importance of a multidisciplinary approach. In this case, the prompt recognition of neurological deterioration allowed for immediate surgical intervention, resulting in complete recovery without sequelae.

INFORMED CONSENT

Written consent obtained from the patient has been collected and preserved by the authors.

ETHICAL APPROVAL

This article dose not contain any studies with human participants or animals performed by any of the authors.

Reference

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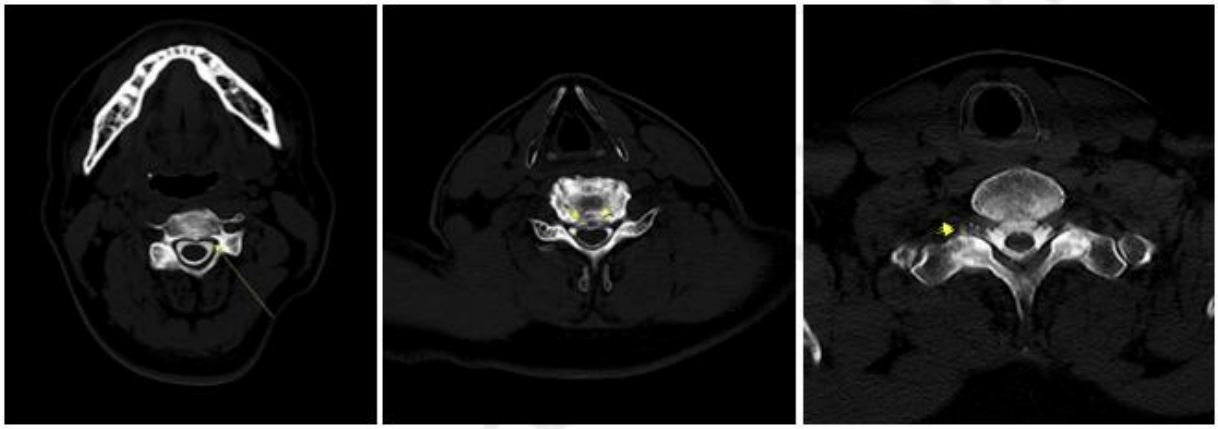

Figure 1.axial CT myelography images, sequentially at the level of C2/3, C5/6, and C7/T1. Yellow arrow indicatescontrast leakage at epidural space.

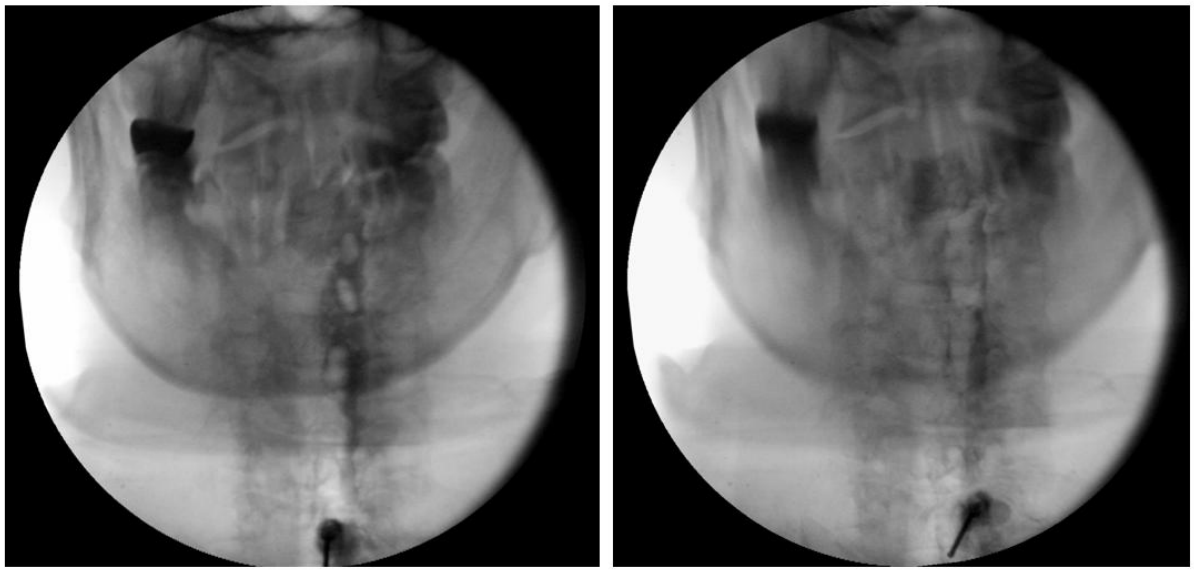

Figure 2. C-arm image of contrast injection during epidural blood patch procedure and C-arm image of contrast diluted by autologous blood injection during blood patch

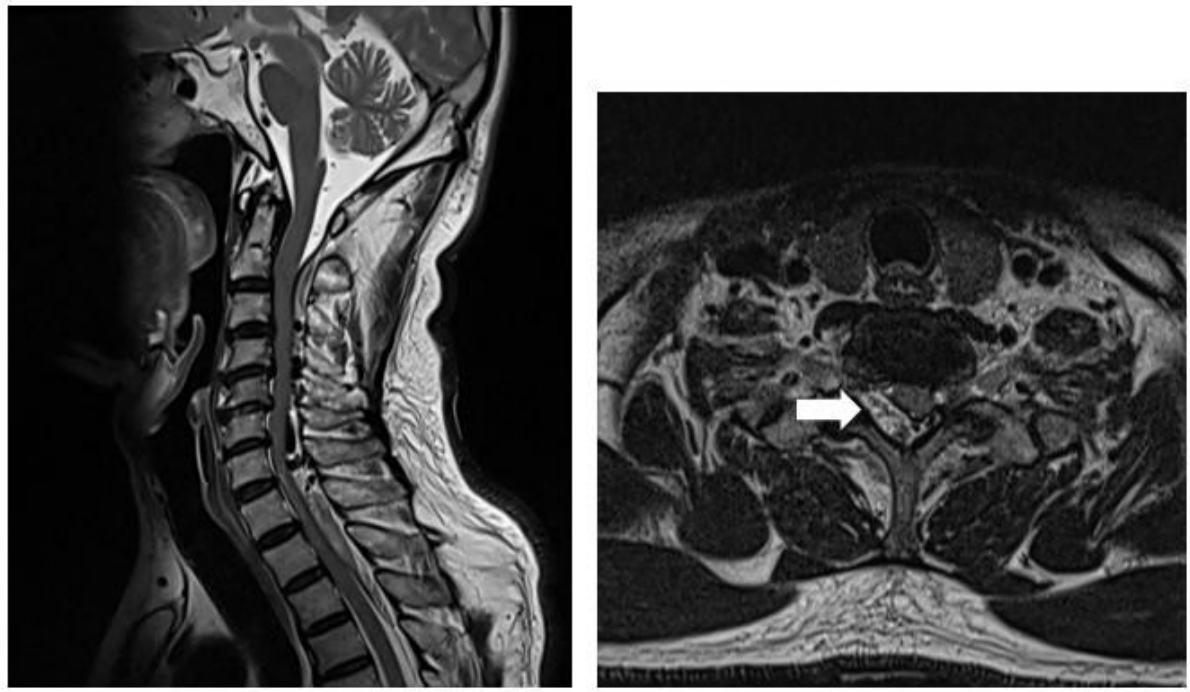

Figure 3.C-spine MRI taken after quadriplegia. A parasagittal T2-weighted image and an axial T2-wighted image at the level of C7/T1. MRI images show epidural hematoma with air bubbles in C2~T4 posterior epidural space.White arrow indicates the externally injected autologous blood may be the cause of cord impingement or compression, C2-3~T2-3.