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Case Report

Epidural Blood Patch Performed for Severe Intracranial Hypotension Following Lumbar Cerebrospinal Fluid Drainage for Intracranial Aneurysm Surgery. Retrospective Series and Literature Review

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Intracranial hypotension (IH) can occur following lumbar drainage for clipping of an intracranial aneurysm. We observed 3 cases of IH, which were all successfully treated by epidural blood patch (EBP). Herein, the authors report our cases.

Keywords Intracranial aneurysm, Intracranial hypotension, Cerebrospinal fluid leakage, Epidural blood patch

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INTRODUCTION

Preceding intracranial aneurysm clipping, a lumbar subarachnoid catheter can be placed to withdraw cerebrospinal fluid (CSF) in order to facilitate surgical exposure. The dural defect created by the lumbar drain commonly seals spontaneously in a few days following catheter removal. Persistent CSF leakage in excess of its production can cause intracranial hypotension (IH) ranging in severity from a mild post-dural puncture headache (PDPH, i.e., headache, neck stiffness, nausea, and dizziness) to severe IH, "brain sag", potentially causing midbrain herniation, pupillary dilation, hemorrhage, lethargy, coma, and death.[13)19)21)24)26)28)] Symptoms usually are exacerbated with sitting and relieved when recumbent.[26)]

Previously, anesthesiologists have been cautioned against performing an epidural blood patch (EBP) in the presence of intracranial pathology.[16)] However, if the intracranial pathology is a direct result of persistent CSF leak, an EBP may be life-saving.[35)]

One of us (P.P.H.) observed that following open repair of anterior circulation aneurysms, some patients were more somnolent than would be anticipated based on surgery itself or other complications (e.g., residual ischemia or vasospasm). Given our height-

Fig. 1. Case 1. A non-contrast head computed tomography reveals left epidural air and fluid collection along the fronto-temporal convexity, slit-like ventricles, and mild left to right midline shift. Not shown is decreased patency of basal cisterns and an oblong midbrain.

ened awareness of possible IH following lumbar drain removal, we monitored clinically for brain sag and employed an EBP when indicated.[2)6)20)27)34)]

We hypothesize that when there is evidence of persistent CSF leakage contributing to brain sag, EBP may be a valuable supplement or alternative to Trendelenburg therapy. Herein, the authors report our cases.

CASE REPORTS

Case 1

A 25-year-old 160 cm tall woman weighing 73 kg had recanalization of a previously coiled anterior communicating artery aneurysm. A Spetzler catheter was inserted at the L4-5 interspace and drained CSF but the volume was not recorded. The aneurysm was successfully clipped and the lumbar drain removed. Postoperatively, she was neurologically intact. On POD 2, she became confused and apathetic; this worsened over the next 3 days. She complained of a positional headache, present while walking or sitting and absent when lying supine, (Fig. 1).

Brain sag was diagnosed by progressive global neurological deterioration, orthostatic headache and computed tomography (CT) findings. On POD 6, an L4-5 EBP was performed with 25 mL of autologous blood. Within several hours, alertness improved, mental status returned to baseline, and headache fully resolved. She was discharged the next day.

Case 2

A 54-year-old 175 cm tall man weighing 84 kg underwent right craniotomy for clipping of an unruptured middle cerebral artery aneurysm. A Spetzler catheter inserted at L4-5 interspace drained 100 mL of CSF, and was removed post-craniotomy. The patient did well initially, however, on POD 3, he developed a partial right third nerve palsy and lethargy. A Head CT revealed midline shift with an epidural air collection (Fig. 2A). Re-exploration was performed to address the mass effect attributed to the epidural collection. At craniotomy, however, the dura and brain were depressed and not under pressure. Postoperatively, upright intracranial pressure (ICP) was -14 mmHg, and increased to 0 with supine positioning. Arteriography demonstrated no vasospasm; although the basilar artery was kinked (Fig. 2B).[2)] A head CT revealed an oval shape midbrain (Fig. 2C), consistent with brain sag. Two days after re-exploration, he was awake but confused, oriented to self, following only simple commands. His right pupil was dilated and nonreactive to light. He remained too lethargic to complain of positional headache but became more alert while supine.

Brain sag was confirmed by the constellation of persistent lethargy, third nerve palsy, radiographic findings and low ICP (0-3 mmHg). Seven days after aneurysm clipping, a L4-5 EBP was performed with 25 mL of blood. Within the hour, he became more awake and appropriate. Within four hours, he en-

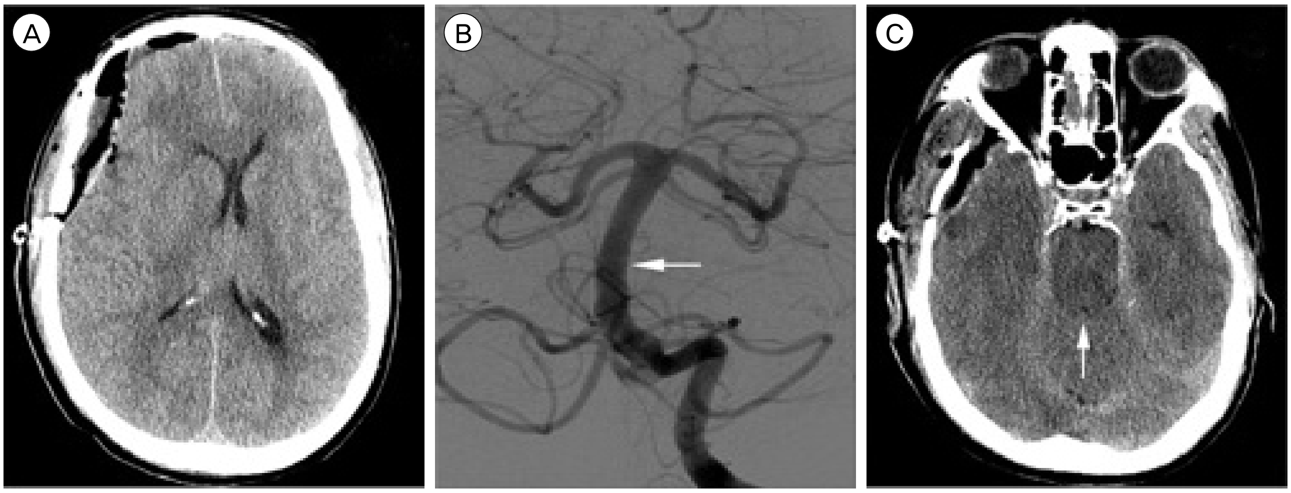

Fig. 2. Case 2. (A) Non-contrast head computed tomography (CT) showing midline shift and an air collection in the right epidural space subjacent to the craniotomy site. (B) This anterior-posterior projection following injection of contrast into the right vertebral artery reveals kinking of the basilar artery above the vertebro-basilar junction (arrow), "cobra sign", and this is consistent with brain sag. (C) Non-contrast head CT reveals an oval shaped midbrain with obliteration of the basilar cisterns (arrow), a sign of brain sag.

gaged in a telephone conversation and fully recovered to baseline neurological state. One day after EBP, he was ready for transfer to rehabilitation medicine and remained neurologically intact thereafter.

Case 3

A 47-year-old 160 cm tall woman weighing 59 kg presented with a ruptured left posterior communicating artery aneurysm, Hunt-Hess Grade 4. She was hemodynamically unstable with pulmonary edema and Takotsubo's cardiomyopathy. A ventricular drain was inserted to relieve hydrocephalus. Following 2 days of medical optimization, the ICP remained normal, she followed commands, and underwent craniotomy for aneurysm clipping. A Spetzler catheter was inserted at the L4-5 interspace. Intraoperatively, significant brain edema was encountered and 50 mL of CSF was drained. After aneurysm clipping, the bone flap was not replaced. The lumbar drain was removed and she remained intubated. Her postoperative head CT showed hemorrhagic contusions of the left frontal and temporal lobes likely due to surgical retraction. On POD 1, her left pupil was dilated and minimally reactive to light. She spontaneously moved her left arm and withdrew on the right. A second Head CT showed diffuse cerebral edema, small lateral ventricles, effacement of the basal cisterns, and herniation of the left cerebellar tonsil through the foramen magnum (Fig. 3). The craniectomy site was soft and the ICP was 6 mmHg.

The neurological and radiological findings in con-

Fig. 3. Case 3. Non-contrast head computed tomography demonstrates herniation of the left cerebellar tonsil through the foramen magnum (arrow).

junction with a soft skin flap and low ICP led to the diagnosis of brain sag. Twenty hours after drain removal, an L4-5 EBP was performed with 20 mL of blood. Within 24 hours, the left pupil returned to baseline, both pupils were reactive, the left arm briskly localized while the right elicited weak extensor posturing, the ICP increased to 12 mmHg, and the craniectomy site had normal tension. She gradually improved, and on POD 16 was transferred to rehabilitation facility. At 6 month follow-up, she had persistent right hemiparesis and breathed spontaneously on a tracheostomy collar.

DISCUSSION

Nine previously reported patients developed brain sag following intracranial aneurysm clipping utilizing a lumbar drain, and were treated with EBP.[2)5)6)27)34)] In 8 of these patients, a rapid, significant, and permanent neurological improvement was reported. The 3 patients in the present series also demonstrated rapid and significant improvement. Overall, EBP improved neurological outcome in 11 of 12 documented cases. One patient with poor outcome after EBP had communicating hydrocephalus, which in conjunction with intraoperative lumbar drainage, caused brain sag and transtentorial herniation leading to intraoperative brainstem infarcts and Duret's hemorrhages.[34)]

Critical IH or "brain sag" was defined by Komotar et al.[22)] as 1) clinical signs of transtentorial herniation (e.g., decline in mental status, pupillary asymmetry or non-reactivity, flexor or extensor posturing), 2) CT imaging revealing an oblong shaped brainstem with effacement of the basal cisterns, and 3) clinical improvement with 15 to 30 degrees of Trendelenburg position. However, brain sag is more precisely defined as "…signs of transtentorial herniation unattributable to other known factors, in association with low intracranial pressure."[3)]

In the 2006 report by the Subarachnoid Hemorrhage Outcomes Project,[23)] brain sag was identified in 16 of 167 patients (9.6%) and was associated with global cerebral edema on initial head CT scan and a longer surgical duration. In the 2005 report by the same group,[22)] the 11 patients who developed brain sag were treated with Trendelenburg position (15 to 30 degrees). Clinical improvement was observed within 12 hours, while sustained symptom resolution required Trendelenburg for an average duration of 2.8 days (± 0.4 days, range 1 to 5 days).[22)]

Treatments for PDPH are well-described.[7)9)17)32)36)] Conservative management including supine bedrest and fluids provide symptomatic relief, but lack consistent long-term benefit.[36)] Injection of autologous blood into the epidural space (EBP) usually provides rapid, effective, and durable symptom relief in the majority of cases.[4)26)32)33)39)] Specific methods, indications, and contraindications for performing an EBP have been reviewed.[7)14)15)26)29)30)32)36)]

Two theories explain how an EBP relieves symptoms of PDPH or IH: 1) injection of epidural blood compresses the lumbar cistern, increasing lumbar CSF pressure, reducing traction on brain structures and 2) clotted blood causes mechanical tamponade, promoting dural sealing.[8)11)14-16)18)33)37)38)] Likely, both mechanisms are important; immediate pain relief is a result of the compressive effect, and the prolonged benefit is due to dural sealing. MRI demonstrates that 20 mL of blood injected into the epidural space disperses approximately 3.5 levels cephalad and 1 level caudad to the site of injection, mostly along the posterior dura, and acts to tamponade the thecal sac.[9)37)40)]

Early literature suggests EBP is successful in 90% of patients.[1)14)16)] In a randomized trial of 42 patients with PDPH following diagnostic lumbar puncture, headache was absent or mild in 95% 24 hours after EBP (15-20 mL), whereas moderate or severe headache was observed in 90% of patients managed without an EBP.[39)] A prospective study of 504 patients with PDPH (after diagnostic lumbar puncture, n = 363; spinal or epidural anesthesia, n = 87; and other spinal injections, n = 54), reported an EBP (mean of 23 ± 5 mL of blood) provided complete or partial relief in 93% of patients.[33)] In a recent review of 29 patients with coma due to IH, an EBP was successful in reversing coma in 85% (6 patients required more than one EBP).[26] If symptoms persist, the EBP can be repeated with improved outcome.[1)4)36]

In the setting of brain sag, despite limited study, it appears reasonable to inject at least 20 mL of autologous blood.[12)31)33)39] Crawford[12] observed that EBP failed to correct PDPH in 17% when the volume of injected blood was less than 20 mL, and that complete relief was observed in 91% of patients when the volume was increased to 20-25 mL. The timing of the EBP also may influence its efficacy; performed within 24 hours after dural puncture, patching was unsuccessful in 71%, whereas after 24 hours it afforded full or partial relief in 96%.[25]

This is a retrospective series and brain sag may have been underdiagnosed. The 3 cases reported were clustered in the last 8 months of the 34 month reporting period, and some earlier patients may have been missed. There was no randomization or control group yet we clearly substantiated the clinical efficacy and expediency of an EBP in the setting of brain sag. It is reasonable to assess brain sag treatments in a larger prospective study.

With regards to patient care, we now employ a more systematic approach to postoperative lumbar drain management to facilitate early diagnosis of IH and to differentiate IH from other causes of neurological decline. We leave the lumbar drain in place for at least one postoperative day. After the patient has fully recovered from general anesthesia and is neurologically stable, the lumbar drain is removed and the patient remains supine. While the patient is being monitored for signs of neurological decline, the head of the bed is elevated to 30°. If there is possible brain sag, the patient is placed supine and observed for signs of clinical improvement. When IH is persistent or severe (i.e., brain sag evident on clinical examination and imaging studies), we more readily perform an EBP. To avoid exacerbating symptoms of IH, we prefer to perform the EBP with the patient in the lateral position.

CONCLUSION

EBP is a reasonable therapeutic option for persistent IH following clipping of an aneurysm, and it usually provides rapid and durable clinical improvement. An EBP should be performed only after thorough patient evaluation and in close collaboration with a neurosurgeon or neurologist. Although performing an EBP in the presence of intracranial pathology may seem counterintuitive, the EBP may be lifesaving in that brain sag may lead to a far worse outcome.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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