계명의대학술지 제30권 2호
Keimyung Med J
Vol. 30, No. 2, December, 2011

Intracranial Hypotension Following Lumbar Drainage

Ealmaan Kim M.D.

Department of Neurosurgery, Keimyung University School of Medicine, Daegu, Korea

Abstract

Lumbar drainage (LD) is a technique that is widely used to prevent cerebrospinal fluid (CSF) leakage following cranial base surgery. Intracranial hypotension (ICH) syndrome with severe neurological symptoms, which is a complication of LD, is a rarely observed entity. In this paper, we present the case of a 51-year-old woman with pituitary macroadenoma, who developed severe ICH after LD and required lumbar intrathecal saline infusion (LISI) in order to reverse the acute obtundation immediately. LISI may be a life-saving intervention in patients with cerebral herniation and coma resulting from CSF depletion secondary to LD that is refractory to conventional treatment modalities.

Key Words : Cerebrospinal fluid leakage, Intracranial hypotension, Intrathecal lumbar infusion, Lumbar drain

Introduction

Although other symptoms exist, intracranial hypotension (ICH) syndrome is mainly characterized by a low opening cerebrospinal fluid (CSF) pressure less than 60 mm H_2O and orthostatic headache [1,2]. ICH syndrome is primary or, more frequently, secondary to anesthetic or therapeutic dural puncture [3]. Most patients of ICH resolve spontaneously with conservative therapy. However, a few may become drowsy, and even lapse into coma, because of a failure to recognize an impending brain herniation [4,5].

Temporary CSF diversion by lumbar drainage (LD) has been commonly used after skull base surgeries in order to prevent postoperative CSF leakage. However, lumbar catheter overdrainage and CSF hypovolemia can result in severe ICH that is manifested by

Corresponding Author: Ealmaan Kim, M.D., Department of Neurosurgery, Keimyung University School of Medicine
56 Dalseong-ro, Jung-gu, Daegu 700-712, Korea
Tel: +82-53-250-7823 E-mail: bach1158@dsmc.or.kr

acute obtundation and dilated, non-reactive pupils, and brain sagging through the tentorial notch and swelling of the diencephalic-mesencephalic structures [6,7]. However, the appropriate management of the obtundation caused by severe ICH is still not well defined. In this paper, we report on a patient who was aged 51 years and who required LD after the endonasal and transcranial surgery for a pituitary tumor. The patient had ICH, which presented initially with headache, and which was followed by a rapidly progressive decline in mental status for which lumbar intrathecal saline infusion (LISI) was necessary to normalize the CSF pressure. This technical note expands the presently known clinical spectrum of ICH, which is typically an uncommon and generally benign illness but which can be a potentially life-threatening condition.

Case Report

A 51-year-old female underwent resection of a giant pituitary macroadenoma by an orbitozygomatic frontotemporal craniotomy after an unsuccessful attempt by a transsphenoidal approach. LD was conducted postoperatively to prevent CSF leakage because of defects in the sellar floor and the suprasellar arachnoid membrane. She recovered uneventfully without additional neurological deficits. Postoperative brain computed tomography (CT) showed minimal changes in the surgical routes and sites. A drainage tube was left open and was initially set to drain CSF continuously by relating the height of the bed. There was no evidence of CSF rhinorrhea.

Early morning of the third day after the operation, she became lethargic and unresponsive with dilated and minimally reactive pupils. Follow-up CT scans showed a large collection of intracranial air and a compression of the brain stem from downward herniation of the brain (Fig. 1A and B). At this point, the pressure of the intrathecal catheter was measured directly with a manometer. The amount of CSF drained on the second day of LD was approximately 350 mL. The negative intrathecal pressure and the CT findings alerted us to the possibility of severe ICH. Thereafter, the lumbar subarachnoid tube was immediately clamped and the head was temporarily placed to the Trendelenberg position. The patient was simultaneously treated with intravenous infusion. Despite these treatments, the patient remained severely obtunded. Therefore, a 30 mL autologous epidural blood patch (EBP) was administered into the lumbar region emergently, and then the patient's neurological picture resolved temporarily within 20 minutes. However, her consciousness stupor returned within 6 hours with hiccups and labored breathing.

Consequently, in order to restore intraspinal and intracranial pressure, immediate consideration was given to LISI, and 50 mL of sterile preservative-free saline was slowly infused back into the lumbar drain over 10 minutes. Shortly thereafter, the patient dramatically awakened and became interactive. The lumbar infusion was continued at 20 mL per hour using an automatic pump. Approximately 8 hours after injecting the saline, the lumbar pressure reached 13 mm H_2O and the patient regained full consciousness.

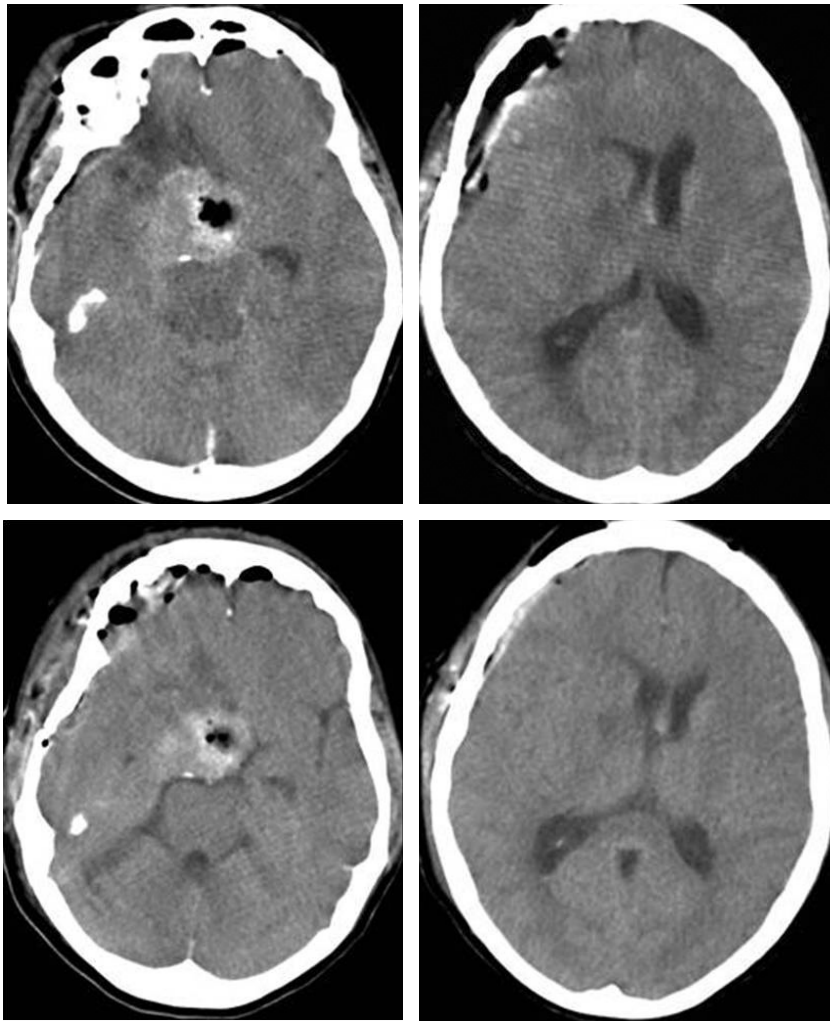

Fig. 1. Computed tomography scans of a patient with pituitary macroadenoma who was operated on through a cranial approach following an insufficient transsphenoidal surgery. This patient developed intracranial hypotension syndrome following lumbar subarachnoid drainage. A, B: Axial images at two levels showing extreme distortion and compression of the brain stem and a large collection of frontotemporal subdural air. The patient was obtunded at this time. C, D: Follow-up images after lumbar intrathecal saline infusion demonstrating complete normalization of the brain stem and basal cisterns.

Radioisotope cisternography performed after the patient was stabilized did not demonstrate any CSF fistula along the neuroaxis. The patient was maintained on bed rest, and then the lumbar catheter was removed on postoperative day 6. In an attempt to seal the possible leak through the lumbar puncture (LP) site, a second EBP was performed. Follow-up CT revealed restoration of the ambient cistern, resolution of the intradural air, and disappearance of the midline shift (Fig. 1C and D). The patient was discharged home in excellent condition.

Discussion

ICH caused by a loss of CSF is an uncommon, but increasingly recognized, neurologic disorder. For diagnostic purposes, the most common cause is anesthetic or therapeutic LP, although spontaneous CSF leakage from a spinal dural defect may occur [4,8]. This syndrome is characterized by brain displacement, subdural collections, dilatated dural venosinuses, pacchymeningeal thickening, and gadolinium enhancement on magnetic resonance images [1,9]. Clinically, lower CSF volume and pressure is typically diagnosed in patients presenting with positional headaches. However, the signs and symptoms can vary according to the brain structure that might be affected during the descent, repositioning, and traction of the anchoring structures. Reports of unusual clinical presentations, such as neck pain, blurred vision, tinnitus, nausea, vomiting, clouding of consciousness, dizziness, and vertigo, continue to appear in the literature [5,7,10]. In addition, because of the considerable variability of the clinical spectrum, a delay in differentiating this condition from disorders that mimic ICH may subject patients to unnecessary procedures and prolong morbidity. Most patients show benign clinical courses, but a severe type of ICH producing acute obtundation has been described in a few reports [11,12]. Theoretically, the mechanism of severe encephalopathy in ICH is presumed to be compression of the diencephalon from downward displacement of the brain [13]. Increasing the awareness of this rarity is the key to the diagnosis of this potentially serious pathological syndrome that can present with orthostatic stupor and coma.

LP or LD is a basic and safe procedure that is highly contributive to neurological diagnosis and management. However, it can also cause serious adverse side effects, including subdural hematomas, pneumocephalus, and ICH, as illustrated by this case report [14]. It has been suggested that persistent CSF leakage at a LP site may cause ICH even in noncomplicated procedures. There was an additional 8% incidence of delayed leak at the site of LP [4,15]. In the case of lumboperitoneal shunt placement, catheter-associated CSF leakage through the hole is an unusual, but important, cause of ICH [16]. For the first time, we have herein described a newly recognized complication of LD, namely, severe ICH from overdrainage of the CSF. Three causative factors may be proposed as a mechanism of ICH from LD following skull base surgery. First, LD promotes ICH with the loss of CSF volume and the downward herniation of brain. Second, air can be drawn into the intracranial space through the skull base defect by negative pressure, further contributing to the downward herniation, particularly in cases involving the anterolateral skull base. This finding was present on CT scans in our case. Last, even after removal of the lumbar catheter, persistent leakage of the CSF can still occur through the dural puncture sites.

External CSF drainage should be meticulously monitored and titrated to control the intracranial pressure while avoiding overdrainage. We have no criteria on CSF drainage volume and rate, however, taking into consideration the normal CSF production rate, approximately 20 mL/h, LD with a draining rate of 30 mL every 4 to 6 hours, total amount of 120 to 180 mL/day, in a bedridden patient, does not produce the

clinical and radiological picture of ICH syndrome [5,17,18]. In addition, LD can be usually continued for 3 to 5 days with a low leak recurrence rate in patients of low-pressure or normal-pressure CSF fistulas. More practically, an accidental siphon effect can be avoided by relating the height of the drainage valve or the drainage bag to the level the ventricular system rather than the bed. In this way, the pressure column remains constant as the bed is raised and lowered or as the patient is moved. A ready-made kits and systems for this purpose allows close regulation of CSF drainage, helps prevent siphoning, and retards retrograde travel of bacteria from the closed collection bottle.

The majority of ICH patients respond well to conservative treatment, but some present stubborn therapeutic challenges. The application of EBP has been proposed as an effective therapy for spontaneous ICH caused by spinal dural tears. Previous estimates of EBP efficacy were overgenerous; persistent symptomatic relief can be expected in more than 80% of patients with initial EBP [15,17]. This epidural intervention can also be repeated in cases of relapse with a high success rate [19]. The primary goal of the patch procedure is to transmit the increased pressure within the epidural space along the entire cord to the CSF space. Its long-term effectiveness is related to dural tamponade or fibrosis that scars over the site of the leak. There is no standard therapeutic strategy for patients who fail to respond to several consecutive blood patches. However, surgical repair or the use of biological glue is sometimes required [20,21]. In order to prevent or treat postdural puncture headache, epidural administration of saline solutions in a continuous or intermittent fashion might be an efficient therapeutic alternative, despite the small number of cases reported in the literature [22]. The injection of normal saline into the subarachnoid space reduces the severity of low CSF pressure headache. This case demonstrates the use of the monitoring of lumbar pressure in order to appropriately titrate the infusion and document the resolution of severe ICH promptly. We have summarized acute management strategies for arresting or reversing an obtundation from brain herniation in the ICH patients. Early recognition and correct diagnosis are crucial for rescuing patients with severe ICH from LD presenting with mental confusion [18]. First of all, clamp the lumbar drain and place the patient's head down, The LD must be kept closed until the patient stabilizes. However, in cases of rapidly declining mental status and impending herniation, saline can be manually or mechanically injected into the lumbar drain in order to expand the CSF volume and normalize pressure [13,23]. Care should be taken to not infuse too quickly in order to prevent a CSF fistula at the closure site and intracranial hypertension. Based on the pressure monitoring and the patient's clinical response, the infusion rate and volume has to be adjusted in order to maintain a target of lumbar intraspinal pressure [24]. Maneuvers aimed at sealing the CSF fistula can then be performed in a less emergent fashion after the patient's mental status has stabilized. If the lumbar drain has been removed and symptoms of ICH recur or persist, an EBP can be considered.

Summary

LD is a versatile and common procedure that is highly contributive to the neurosurgical management of patients with CSF fistulae after skull base surgery. Rarely, it may cause serious adverse effects, namely, severe ICH occurring from CSF overdrainage, as illustrated by this report. Although it is very unusual for severe ICH to present with coma, it is important to recognize that a coma that does not resolve with conservative therapies can be reversed with the LISI technique.

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