

자발성 두개 내 저혈압의 뇌조조영술 소견

아주대학교 의과대학 핵의학교실, 신경과학교실¹

김수지 · 박찬희 · 배문선 · 윤석남 · 오윤민¹ · 김장성¹

Evaluation of Spontaneous Intracranial Hypotension Using Radionuclide Cisternography

Suzy Kim, M.D., Chan H. Park, M.D., Moonsun Pai, M.D., Seok-nam Yoon, M.D.,
Yunmin Oh, M.D.¹ and Jangsung Kim, M.D.¹

Departments of Nuclear Medicine and Neurology¹, Ajou University School of Medicine, Suwon, Korea

Abstract

We report four cases of spontaneous intracranial hypotension that were investigated by radionuclide cisternography. ^{99m}Tc-diethylenetriamine pentaacetic acid radionuclide cisternography of all our patients showed direct sign of cerebrospinal fluid leakage as well as indirect signs of less activity than expected over the cerebral convexities and rapid appearance of bladder activity. The headache of all patients was eventually controlled with bed rest and hydration. (*Korean J Nucl Med* 1999;33:178-83)

Key Words: Spontaneous intracranial hypotension, Cerebrospinal fluid, Radionuclide cisternography, Tc-99m DTPA

Introduction

Spontaneous intracranial hypotension (SIH) is characterized by postural headache in conjunction with intracranial hypotension and typically occurs without an obvious causes such as lumbar puncture, craniotomy, other operative procedures, traumatic dural tear, or other medical conditions.¹⁾ However, it can be seen following the sneezing, coughing, intercourse or minor fall.^{1,2)} Spontaneous

spinal cerebrospinal fluid (CSF) leaks are not common, but are now increasingly recognized as a cause of postural headache associated with intracranial hypotension.³⁾ The purpose of our study was to describe different clinical and radiological features of four SIH cases and also to evaluate the role of radionuclide cisternography in the diagnosis of SIH caused by spinal CSF leakage.

Case Reports

1. Patient 1

A 66-year-old female patient developed sudden onset of severe headache while sitting and standing for 1 week duration. Ten days earlier she had symptoms of upper respiratory infection. She had a

Received Oct. 27, 1998; revision accepted Mar. 20, 1999
Corresponding Author: Chan H. Park, M.D., Department of Nuclear Medicine, Ajou University Hospital, San 5, Wonchun-dong, Paldal-gu, Suwon 442-749, Korea
Tel: (0331) 219-5847/8, Fax: (0331) 219-5950
E-mail: chpark@madang.ajou.ac.kr

past history of tympanic membrane rupture 3 years ago. The headache was worsened by the upright position and relieved in supine position within 30 minutes. The headache was associated with nausea, dizziness, and eyeball pain. Physical and neurologic examinations were unremarkable except for neck stiffness. Routine laboratory findings were also normal. Examination of CSF showed pressure of 30~40 mmH₂O (N: 50~180 mmH₂O), WBC of 5/ml (N:<5/ml), glucose of 65 mg/dL (N: 40~70 mg/dL) and protein concentration of 44 mg/dL (N: 20~50 mg/dL). Brain magnetic resonance imaging (MRI) revealed mild dural thickening, mild suprasagittal vein engorgement and diffuse pachymeningeal gadolinium enhancement (Fig. 1A). There was no mass effect and no tonsillar herniation. Radionuclide cisternography was performed after injection of 37~74 MBq of ^{99m}Tc-diethylenetriamine pentaacetic acid (DTPA) into the lumbar subarachnoid space and the images were obtained at 1, 3, 6, 24 hours using a single head gamma camera equipped with low energy all purpose collimator. Entire spinal region in posterior view and head in frontal and lateral views were obtained. The scan depicted early renal excretion with out-pouching of lumbar subarachnoid space along the nerve roots as well as at the cervicothoracic junction on the right side (Fig. 1B). MRI of cervico-thoracic spine performed 16 days after the onset of the headache showed no abnormal finding. After treatments with intravenous hydration and analgesics, the patient's headache was completely subsided.

2. Patient 2

A 32-year-old female patient was hospitalized for persistent headache accompanied by transient episodic tinnitus for at least 10 days duration. She had no history of trauma, operation or medical illness. The headache was aggravated by standing and disappeared when she was in recumbent posi-

tion. On admission, physical and neurologic examinations were normal. Lumbar puncture was difficult to obtain. There was no spontaneous flow and it was necessary to aspirate CSF. Examination of CSF showed glucose level of 58 mg/dL and elevated protein concentration of 178 mg/dL. Brain MRI revealed dural thickening along the falx and tentorium as well as leptomeningeal enhancement on the gadolinium enhanced T1 weighted images. Radionuclide cisternography was performed using ^{99m}Tc-DTPA and demonstrated a large extradural leakage of CSF at cervicothoracic junction and multiple small foci of CSF leakage at thoracic

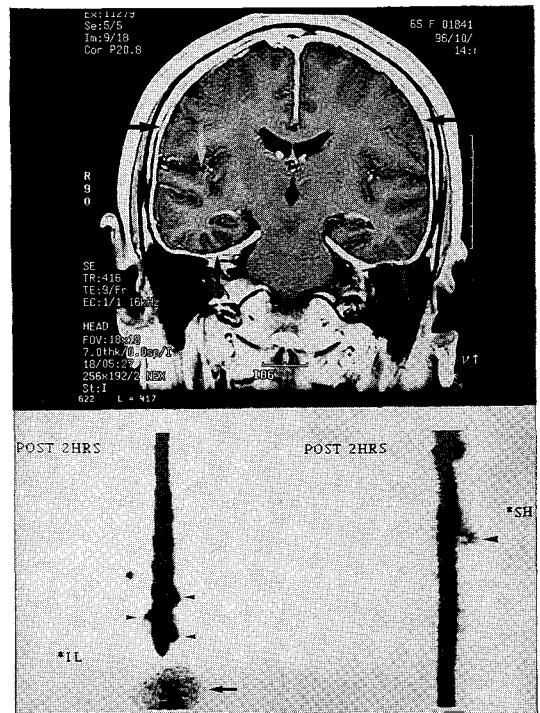


Fig. 1. Brain MRI of 66-year-old woman with spontaneous intracranial hypotension depicts mild dural thickening, mild suprasagittal vein engorgement and diffuse pachymeningeal enhancement (arrows)(1A). ^{99m}Tc-DTPA RN cisternography shows multiple areas of CSF leakage at cervicothoracic junction and lumbar region (arrows) with early visualization of the bladder(1B). (IL; ilium)

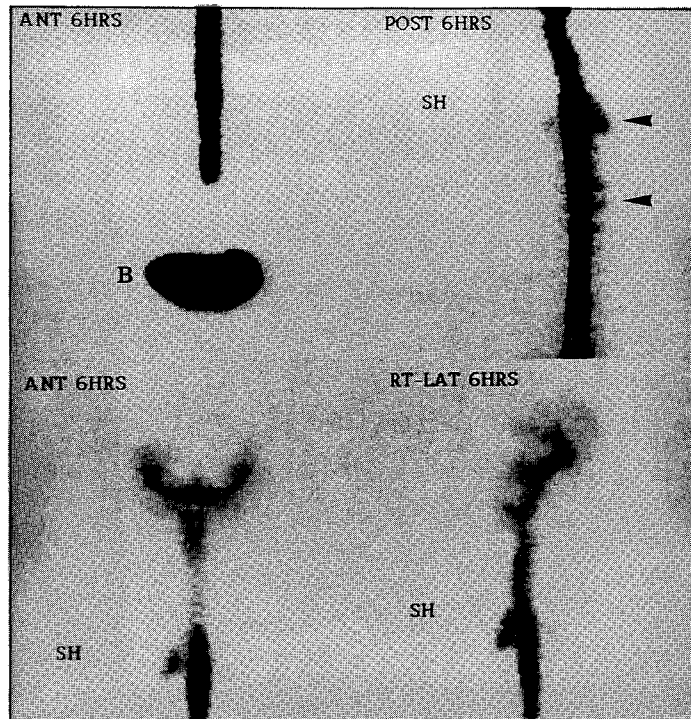


Fig. 2. Radionuclide cisternography of 32-year-old female who complained of persistent headache accompanied by transient episodic tinnitus. At 6 hrs after the injection of ^{99m}Tc -DTPA, multiple foci of CSF leakage at cervicothoracic junction and thoracic region (arrows) are shown (B; bladder SH; shoulder).

region. There was a slow ascent of CSF along the spinal axis as well as early appearance of the bladder activity (Fig. 2). In order to evaluate any organic abnormality, MRI of C-spine was performed. But, the MRI finding was unremarkable. One week after the treatment with oral and intravenous hydration, the patient showed improvement of the headache. Then she was discharged from the hospital to be followed at the outpatient clinic.

3. Patient 3

A 32-year-old male patient presented with a five day history of headache. The headache localized to the frontal area developed right after standing and walking. Physical and neurologic examinations were normal. Lumbar puncture showed a CSF

pressure of 20 mmH₂O, WBC of 4/ml, protein concentration of 43 mg/dL, and glucose level of 61 mg/dL. Routine blood tests were normal as was the brain computed tomography (CT). Brain MRI with gadolinium enhancement demonstrated diffuse pachymeningeal enhancement over the bilateral cerebral and cerebellar hemispheres. ^{99m}Tc -DTPA radionuclide cisternography showed a large area of CSF leakage at cervicothoracic junction (Fig. 3) and early visualization of the bladder. The headache was completely resolved only with oral and intravenous hydration.

4. Patient 4

A 35-year-old male patient experienced orthostatic headache of abrupt onset the day before admis-

sion. Physical and neurologic examinations were unremarkable as was the routine laboratory tests. Lumbar puncture revealed uncheckable CSF pres-

sure. Brain MRI with gadolinium enhancement showed no definite pachymeningeal enhancement. However, radionuclide cisternography demonstrated multiple foci of CSF leakage at cervico-thoraco-lumbar regions and early excretion of the tracer through the kidney into the bladder (Fig. 4). Bed rest and hydration made him to be free from the headache.

Discussion

Spontaneous intracranial hypotension is characterized by headache and low CSF pressure and SIH was first described by Schaltenbrand in 1938.⁴⁾ In this article, the author proposed three possible mechanisms to explain the pathophysiology of the syndrome: 1) decreased production of CSF by choroid plexi; 2) hyperabsorption of CSF by arachnoid villi; 3) small dural tears leading to CSF leakage.

The clinical presentation of the syndrome is rather typical and all our four patients showed typical features of SIH. The headache in SIH is probably due to dilatation of the cerebral veins and meningeal vasculature as there is a reciprocal rela-



Fig. 3. ^{99m}Tc-DTPA Radionuclide cisternography of 32-year-old female with headache reveals a large area of CSF leakage at cervicothoracic junction (arrow)(SH; shoulder).

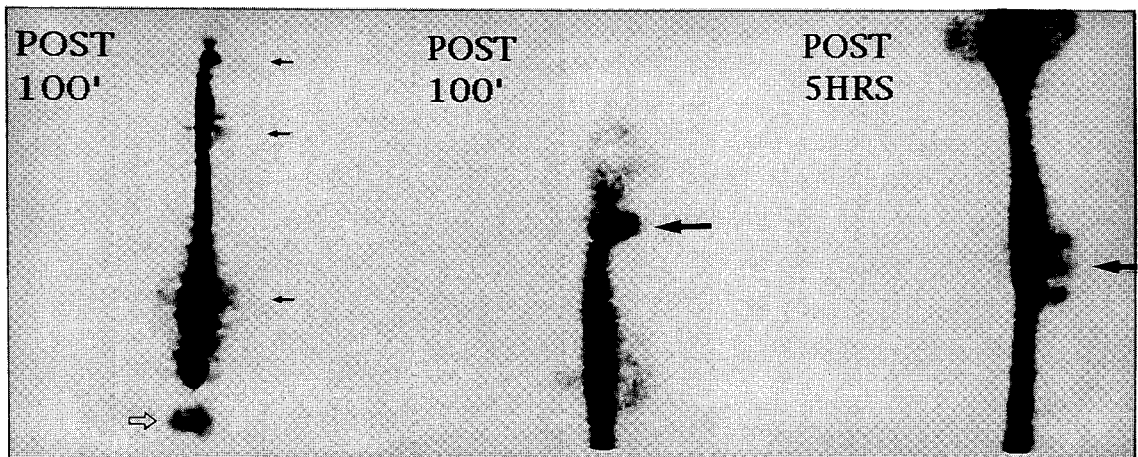


Fig. 4. Radionuclide cisternography of 35-year-old male shows early renal excretion (open arrow) and CSF leakage at cervico- thoraco-lumbar regions (arrows).

tion between CSF volume and intracranial blood volume.⁵⁾ Alternatively, the headache may be due to downward sagging of the brain with traction of the pain sensitive structures when the patient is in upright position. Or it may be caused by both factors.⁶⁾ Nausea, vomiting, tinnitus, vertigo, photophobia, and photophobia are commonly associated with SIH.⁷⁻⁹⁾

The diagnosis can be suggested by history and lumbar puncture. Lumbar puncture is often difficult to obtain in SIH. CSF pressure usually ranges from 0 to 30 mmH₂O.¹⁰⁾ As the spinal fluid pressure falls, the normal hydrostatic and oncotic pressures across the venous sinus and arachnoid villi are disrupted. Perhaps this mechanism explains the moderate elevation of spinal fluid protein.¹⁰⁻¹²⁾ The major abnormalities in brain MRI seen in patients with SIH are the diffuse thickening of the meninges and choroid plexi.^{13,14)}

Once the diagnosis of intracranial hypotension is established, further investigation is necessary to determine a possible cause.¹⁵⁾ Our patients had no history of head or back trauma, recent operative procedure including lumbar puncture, or associated medical illness that could account for intracranial hypotension. Meningitis and CNS malignancies were essentially ruled out with negative CSF cytologies and negative CT or MRI.

The possibility of spontaneous CSF leak was investigated by radionuclide cisternography. Radionuclide cisternography has been shown to be fairly characteristic in patients with SIH, showing early accumulation of radioactive tracer in the bladder and less activity than expected over the cerebral convexities suggesting rapid uptake of tracer into the bloodstream. This is probably due to rapid absorption into the epidural venous plexus.^{1,3,16-20)} This abnormal pattern, however, does not necessarily differentiate between a CSF hyperabsorption and a CSF leak. In some cases a leak

along the spinal axis has been clearly identified, while in others it has not. In the latter situation, this may be due to rapid absorption of the tracer or the leak is not contained. This may also be due to the fact that resolution of the scan is not adequate to demonstrate leakage.^{1,3,16,17)}

Most spinal CSF leaks are located at the cervicothoracic junction or in the thoracic spine. A CSF leak can be detected directly by accumulation of radioactivity outside the subarachnoid space or suggested indirectly by the rapid disappearance of tracer from the subarachnoid space and early appearance in the urinary bladder. In previous reports, most patients with SIH showed indirect evidence of CSF leak only.²¹⁾ However, both indirect and direct radionuclide cisternographic signs described above were present in all of our four patients. We may suggest that different imaging methods or patient population explain the difference. Standardization of imaging protocol or inclusive repeated imaging of thoracolumbar spine including anterior and lateral view. It is not clear. We do not know the exact reason of. In this paper we present two unusual cases of CSF leak identified by radiopharmaceutical cisternogram.

Some authors believe that radiographic study of choice in SIH is CT myelography. Myelography with subsequent CT scanning through the area suspected harboring a CSF leak is sometimes required to localize the leak more precisely. This may reveal an underlying anatomical defect causing the leak such as a meningeal diverticulum. However, it is a painful invasive procedure. If it is performed without a previous imaging study that demonstrates an approximate location of the leak, then thin CT slices should be obtained from the skull base to distal lumbar region.

We concluded that radionuclide cisternography is a useful method to evaluate patients with postural headache due to SIH. Our studies demon-

strated direct sign of CSF leakage in all patients.

References

- 1) Renowden SA, Gregory R, Hyman N, Hilton-Jones D. Spontaneous intracranial hypotension. *J Neurol Neurosurg Psychiatry* 1995;59:511-5.
- 2) Vilaning ST, Titus F. Low cerebrospinal fluid pressure. In: Olesen J, Tfelt-Hansen P, Welch KMA, editors. *The headaches*. New York: Raven Press; 1993. P. 687-95.
- 3) Schievink WI, Meyer FB, Atkinson JLD, Mokri B. Spontaneous spinal cerebral fluid leaks and intracranial hypotension. *J Neurosurg* 1996;84:598-605.
- 4) Schaltenbrand G. Neure anschauen zor pathophysiologie der liquorzirkulation. *Zenfrabl Nforchir* 1938;3:290-300.
- 5) Burrow B. On disorder of the cerebral circulation. London 1846. From Lundberg N. The saga of the Monro-Kellie doctrine. In: Ischie S, Nagai H, Brocks M. *Intracranial pressure V*. Berlin: Springer-Verlag;1983:68-75.
- 6) Wolff HG. *Headaches and other head pain*. 2nd ed. New York: Oxford University Press;1963:96-125.
- 7) Lasater GM. Primary intracranial hypotension. *Headache* 1970;10:63-6.
- 8) Bell WE, Joynt RJ, Sahs AL. Low spinal fluid pressure syndromes. *Neurology* 1961;10:512-21.
- 9) Shenkin HA, Finneson BE. Clinical significance of low cerebral spinal fluid pressure. *Neurology* 1958;8:157-63.
- 10) Cass W, Edelist G. Postspinal headache. *JAMA* 1974;227:786-7.
- 11) Peterson RC, Freeman DP, Knox CA, Gibson BE. Successful treatment of spontaneous low cerebrospinal fluid pressure headache. *Annals of Neurology (Abstract)* 1987;22:148.
- 12) Sipe IC, Zyroff J, Waltz TA. Primary intracranial hypotension and bilateral isodense subdural hematomas. *Neurology* 1981;31:334-7.
- 13) Pannullo SC, Reich JB, Krol G, Deck MDF, Posner JB. MRI changes in intracranial hypotension. *Neurology* 1993;43:919-26.
- 14) Fishman RA, Dillon WP. Dural enhancement and cerebral displacement secondary to intracranial hypotension. *Neurology* 1993;43:609-11.
- 15) John M, Stephen DS. Spontaneous low cerebrospinal fluid pressure headache. *Headache* 1990;30:192-6.
- 16) Benamor M, Tainturier C, Graveleau P, Pierot L. Radionuclide cisternography in spontaneous intracranial hypotension. *Clin Nucl Med* 1998;23:150-1.
- 17) Schievink WI, Reimer R, Folger WM. Surgical treatment of spontaneous intracranial hypotension associated with a spinal arachnoid diverticulum (Case report). *J Neurosurg* 1994;80:736-9.
- 18) Weber WEJ, Heidendal GAK, Dekrom MCTFM. Primary intracranial hypotension and abnormal radionuclide cisternography. *Clin Neurol Neurosurg* 1991;93:55-60.
- 19) Molins A, Alvarez J, Sumalla J. Cisternographic pattern of spontaneous liquoral hypotension. *Cephalalgia* 1990;10:59-65.
- 20) Labadie EL, van Antwerp J, Bamford CR. Abnormal lumbar isotope cisternography in an unusual case of spontaneous hypoliquorrheic headache. *Neurology* 1976;26:135-9.
- 21) Ali SA, Cesani F, Zuckermann JA, Nusynowitz ML, Chaljub G. Spinal-cerebrospinal fluid leak demonstrated by radiopharmaceutical cisternography. *Clin Nucl Med* 1998;23:152-5.