Scientific Research Publishing

Open Journal of Modern Neurosurgery, 2022, 12, 222-232
https://www.scirp.org/journal/ojmn
ISSN Online: 2163-0585
ISSN Print: 2163-0569

# Recurrent Intracranial Hypotension Complicated with Subdural Hematoma—A Case Report and a Case Series Review

**Chi-Man Yip**

Division of Neurosurgery, Kaohsiung Veterans General Hospital, Taiwan, China
Email: yip_chiman@yahoo.com

**How to cite this paper:** Yip, C.-M. (2022) Recurrent Intracranial Hypotension Complicated with Subdural Hematoma—A Case Report and a Case Series Review. *Open Journal of Modern Neurosurgery*, **12**, 222-232.
https://doi.org/10.4236/ojmn.2022.124024

**Received:** September 26, 2022
**Accepted:** October 21, 2022
**Published:** October 24, 2022



## Abstract

Intracranial hypotension has variable clinical manifestations; subdural hematoma is one of the complications of intracranial hypotension with the reported incidence ranging from 16% to 57%. The author would like to share a case of subdural hematoma caused by recurrent intracranial hypotension with different cerebrospinal fluid (CSF) leakage site and to review a case series of intracranial hypotension treated in the author's hospital (Kaohsiung Veterans General Hospital). A 44-year-old male having the past history of intracranial hypotension was treated in our Neurology division one month previous to this admission, who was sent to our emergency room (ER) due to severe orthostatic headache with nausea and vomiting. Computed tomography (CT) scan of brain at ER showed bilateral subdural hematoma, more on the left side with mass effect. Both surgical removal of the subdural hematoma and epidural blood patch were performed and he had a good outcome. Orthostatic headache is a specific symptom sign of intracranial hypotension. Epidural blood patch is effective to manage intracranial hypotension either the leakage site of CSF is detected or not. In case of non-traumatic subdural hematoma, intracranial hypotension should be kept in mind.

## Keywords

Epidural Blood Patch, Intracranial Hypotension, Orthostatic Headache, Subdural Hematoma

## 1. Introduction

Intracranial hypotension occurs when imbalance in the production, absorption, or flow of cerebrospinal fluid (CSF) leads to low intracranial pressure and sagging of the brain within the skull [1]. Intracranial hypotension most com-

monly occurs from a persistent CSF leak after spinal tap but may also be spontaneous [2] [3]. The estimated incidence of spontaneous intracranial hypotension is five cases per 100,000 person-years and female is more often affected [4] [5]. The exact etiology of spontaneous spinal CSF leaks is not certain, but probably caused by the weakness of the meninges surrounding the spinal cord [3] [6] [7]. An underlying connective tissue disorder such as Marfan syndrome, Ehlers-Danlos syndromes, autosomal dominant polycystic kidney disease, neurofibromatosis type 1, and Lehman syndrome may result in dural weakness and play a role in the development of spontaneous intracranial hypotension [3] [5] [8].

Orthostatic headache is a characteristic symptom sign, other common clinical presentations include dizziness or vertigo, nausea and vomiting, tinnitus, changes in hearing, cranial neuropathies, disequilibrium, and visual disorder [1] [2] [3] [5]. The reported incidence of subdural hematoma among spontaneous intracranial hypotension ranges from 16% to 57% [9]. The management of subdural hematoma related to intracranial hypotension includes conservative therapy alone, epidural blood patch with or without surgical removal of the subdural hematoma [9].

The author would like to share a case of subdural hematoma caused by recurrent intracranial hypotension and to retrospectively review a case series of intracranial hypotension treated in the author's hospital.

## 2. Case Report

A 44-year-old male without any systemic disease, previous surgical event and traumatic event who came to our ER on 8th December 2021 due to orthostatic headache, nausea, vomiting, photophobia and neck stiffness for 3 days. On his arrival at our ER, his recorded Glasgow Coma Scale was E4V5M6; his vital signs were stable but presented with acute ill-looking. Brain computed tomography (CT) was checked showing neither intracranial hemorrhage nor space occupying lesion. He was admitted to our Neurology ward for further evaluation and management.

After admission to our Neurology ward, brain magnetic resonance imaging (MRI) was arranged which demonstrated diffuse pachymeningeal dural enhancement over the bilateral fronto-temporo-parieto-occipital, falx and tentorial regions, and mild prominence of the pituitary gland and venous sinuses (**Figure 1**). Spontaneous intracranial hypotension was diagnosed based on the clinical features and the brain MRI findings. Whole spine MRI and magnetic resonance myelography was checked which revealed CSF leakage at the right aspect of dural sac at the T5 level (**Figure 2**). Epidural blood patch at the T5 level was performed by anesthesiologist. After the epidural blood patch procedure, fluid hydration and bed rest, his symptoms signs were resolved and he was discharged home on 17th December 2021.

This patient came back to our ER again on 31st January 2022 with the chief

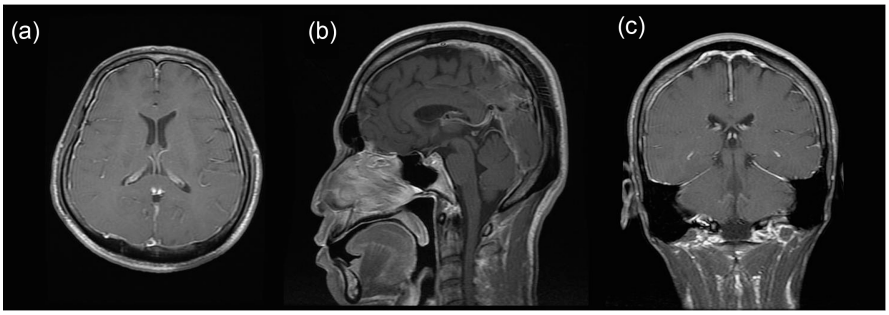


**Figure 1.** Brain magnetic resonance images obtained from the patient before treatment in the first admission. (a) axial post-Gadolinium T1-weighted image of the brain demonstrating intense dural enhancement along the cranial convexity. (b) sagittal post-Gadolinium T1-weighted image of the brain demonstrating intense dural enhancement along the clivus and enlargement of the pituitary gland. (c) coronal post-Gadolinium T1-weighted image of the brain showing intense pachymeningeal enhancement over the convexity and along the tentorium.

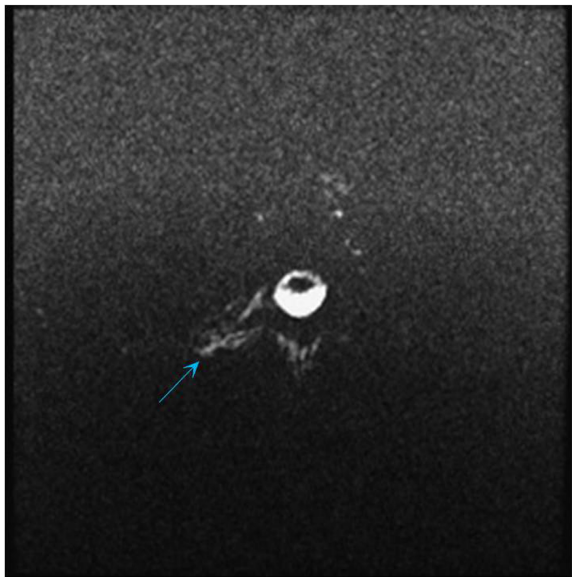

**Figure 2.** Whole spine magnetic resonance myelography obtained from the patient before treatment in the first admission. The arrow pointed out the CSF leakage at the right aspect of dural sac, T5 level.

complaint of orthostatic headache, nausea, vomiting for one day. Cranial computerized tomography was repeated which showed bilateral subdural hematoma, more on the left side with mass effect and midline deviation to the right side (**Figure 3**). He and his family reported no traumatic event and his laboratory data were within normal limits. Urgent burr hole drainage of the left subdural hematoma was performed by the author to release the mass effect and further investigation of the subdural hematoma development was arranged after the surgery. Since he just had the diagnosis and treatment of spontaneous intracranial hypotension, CSF leakage at the T5 level related; residual or recurrent CSF leakage with resultant intracranial hypotension was highly suspected to be

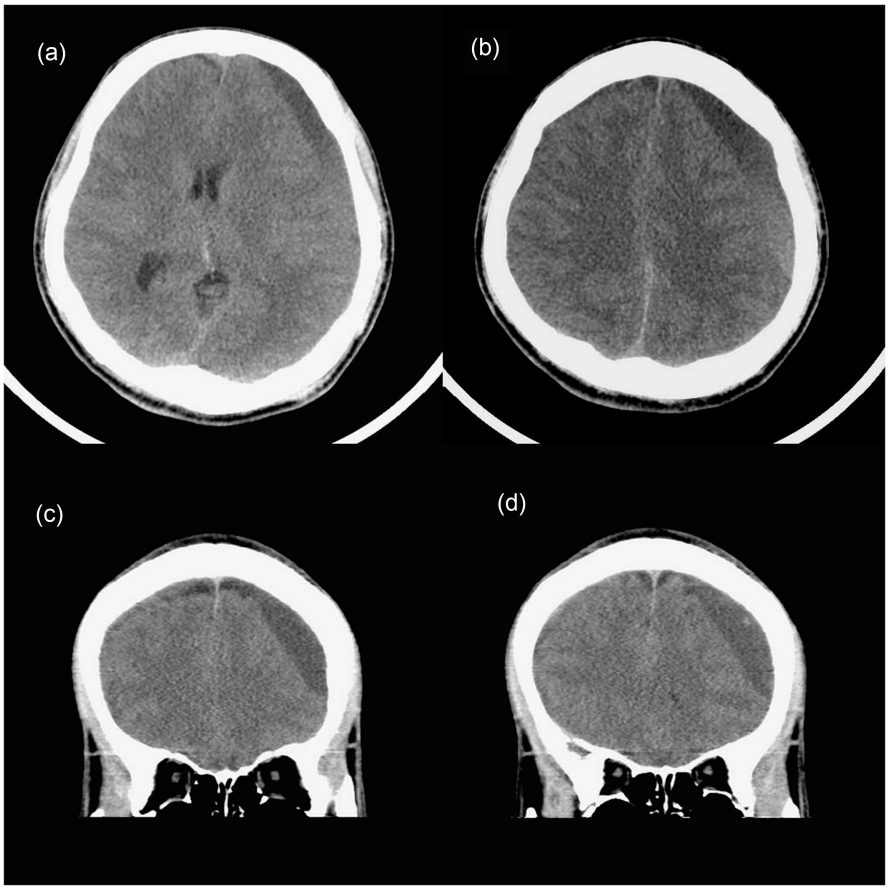


**Figure 3.** Noncontrast cranial computerized tomography at ER. (a) and (b) axial views of brain CT showing iso- to low density subdural hematoma in the left fronto-temporo-parietal-occipital regions, with mass effect on the brain parenchyma. (c) and (d) coronal views of brain CT showing isodensity subdural hematoma in the right fronto-parietal region.

the etiology of his subdural hematoma.

Whole spine MRI was repeated, which showed no evident CSF leakage at the T5 level but suspicious CSF leakage at the right aspect of dural sac at the L2 level (**Figure 4**). Epidural blood patch at the L2 level was performed again by anesthesiologist. Post-operative brain CT demonstrated the removal of the left subdural hematoma and less amount of the right subdural hematoma (**Figure 5**). After the burr hole drainage of the subdural hematoma, the epidural blood patch procedure, fluid hydration and bed rest, his symptoms signs were resolved and he was discharged home on 12th February 2022. He was back to work in March 2022. In June 2022, brain MRI was followed which showed the regression of diffuse pachymeningeal enhancement over the bilateral fronto-temporo-parieto-occipital, falx and tentorial regions (**Figure 6**). However, what caused him to suffer from the CSF leak in different sites remained unknown.

## 3. Materials and Methods

The author searched the keyword "headache" in the discharge summary of all

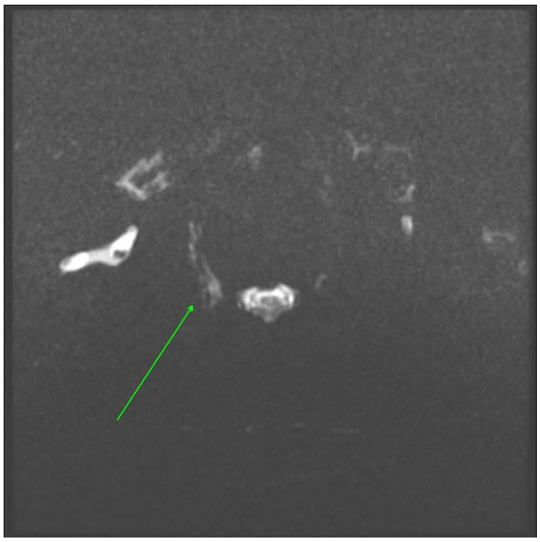

**Figure 4.** Whole spine magnetic resonance myelography obtained from the patient before treatment in the second admission. The arrow pointed out the CSF leakage at the right aspect of dural sac, L2 level.

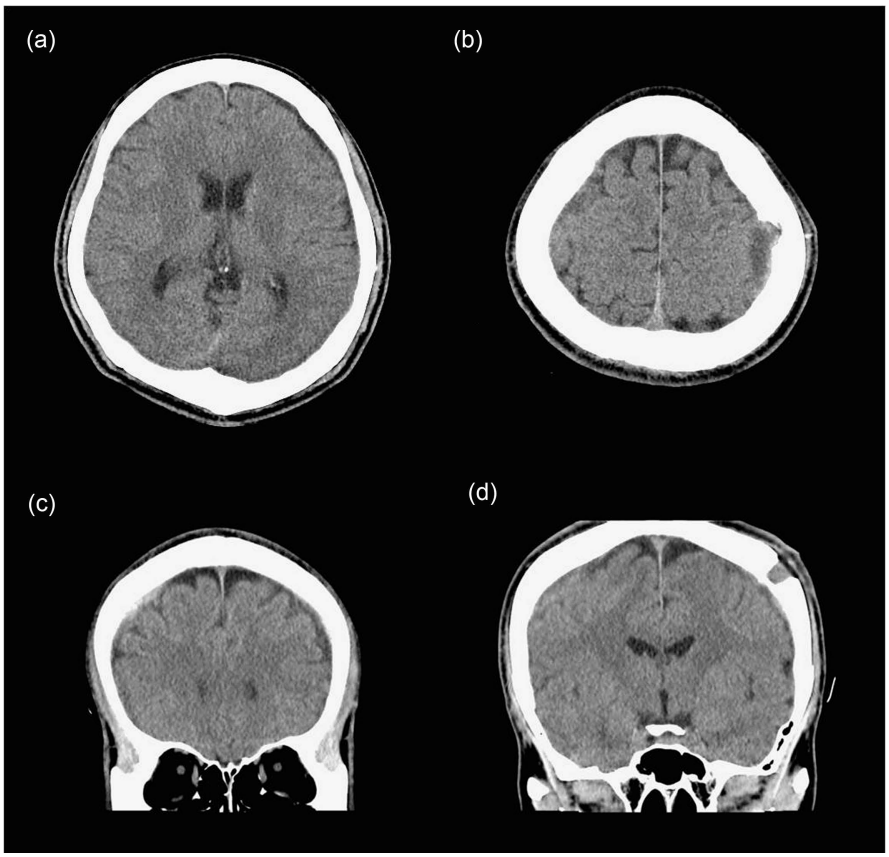


**Figure 5.** Postoperative noncontrast cranial computerized tomography. (a) and (b) axial views of brain CT showing post left high parietal burr hole craniostomy with the removal of subdural hematoma in the left fronto-temporo-parietal-occipital regions, no obvious mass effect. (c) and (d) coronal views of brain CT showing regression of subdural hematoma in the right fronto-parietal region.

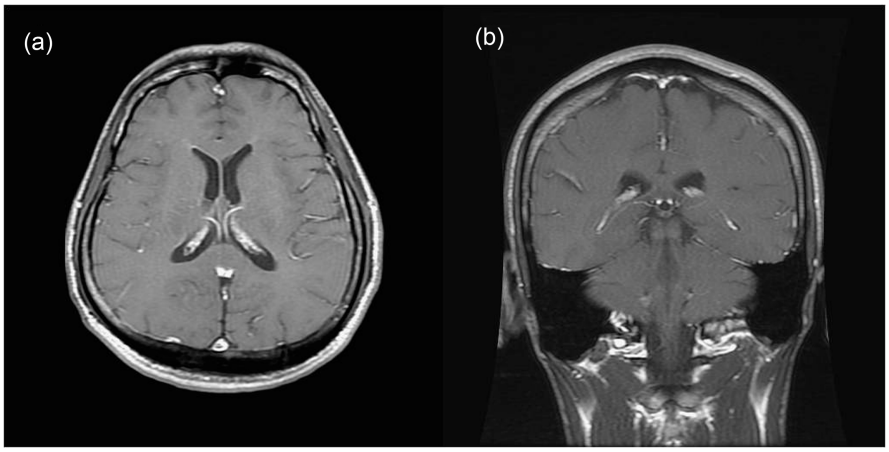


**Figure 6.** Follow-up brain magnetic resonance images 4 months after final treatment. (a) axial post-Gadolinium T1-weighted image of the brain, (b) coronal post-Gadolinium T1-weighted image of the brain demonstrating regression of diffuse pachymeningeal enhancement noted over the bilateral fronto-temporo-parietal, falx and tentorial regions.

patients from June 2012 to February 2022 and excluded those were not diagnosed as intracranial hypotension. The author conducted a retrospective review of the medical records of patients having intracranial hypotension, who were treated in our hospital between June 2012 and February 2022. Their age at diagnosis, sex, clinical presentation, brain MRI findings, spine MRI findings, CSF leakage site, management and associated complication were reviewed and analyzed.

## 4. Results

There were 27 consecutive patients having intracranial hypotension treated in the author's hospital between June 2012 and February 2022. Twelve of them (44.44%) were male and 15 patients (55.56%) were female with the male to female ratio 1:1.25. Their age at diagnosis ranged from 21-year-old to 63-year-old with a mean age of 40.85 years. Their clinical presentations include orthostatic headache, dizziness/vertigo, nausea, vomiting, photophobia, neck stiffness, neck pain, back pain, phonophobia and tinnitus. 26 patients (96.29%) had checked spine MRI and MR myelography while 24 patients (88.89%) had finished the brain MRI. Among these 24 patients who had done brain MRI, 17 patients (70.83%) showed pachymeningeal enhancement; 4 patients (16.67%) showed engorgement of intracranial venous sinus; 3 patients (12.5%) showed enlargement of the pituitary gland; 3 patients (12.5%) showed downward displacement of the brain and 1 patient (4.17%) showed subdural effusion; 5 patients (20.83%) showed negative findings. For those patients who had checked spine MRI and MR myelography (26 patients), CSF leakage was not detected in 7 patients (26.92%); CSF leakage was demonstrated but the CSF leakage site cannot be identified precisely in 6 patients (23.08%); CSF leakage site can be identified in 13 patients (50%). The most frequent site of CSF leakage was T-spine followed by C-spine and then L-spine.

All patients had been treated by fluid hydration and bed rest. 8 patients (29.63%) did not undergo epidural blood patch procedure; 2 of them had CSF leakage shown on the spine MRI, but the definite leakage site could not be identified. 19 patients (70.37%) had undergone epidural blood patch management; 2 of them underwent epidural blood patch management even CSF leak was not detected on the spine MRI. 5 patients underwent twice epidural blood patch procedures. All epidural blood patch procedures were performed by our anesthesiologists. All patients got the relief of their symptoms after either conservative treatment or epidural blood patch management.

Among 27 patients, 3 patients (11.11%) were associated with subdural hematoma; all of them had undergone epidural blood patch management and 2 of them (case #10 and #25) needed surgical intervention to the subdural hematoma. The demographic and clinical characteristics of the 27 patients were summarized in Table 1.

## 5. Discussion

CSF leak is the most common cause of intracranial hypotension and orthostatic headache is a characteristic symptom sign of intracranial hypotension [2] [3]. Persistent CSF leakage leads to CSF hypovolemia, low intracranial pressure and sagging of the brain within the skull. The brain "sags" in the cranial cavity, causing traction on the anchoring and supporting structures of the brain, pain-sensitive intracranial and meningeal structures, and bridging veins, is thought to cause headache and some of the associated symptoms of intracranial hypotension [1] [2] [6]. The postural component of the headache is attributed to accentuated traction in the upright position due to gravity [2]. Secondary vasodilation of the cerebral veins to compensate for the low CSF pressure may contribute to the vascular component of the headache by increasing brain volume [8]. The precise cause of spontaneous spinal CSF leak remains uncertain, but an underlying structural weakness of the spinal meninges is highly suspected to be related [3] [6] [7]. Therefore, general generalized connective tissue disorders, such as Marfan syndrome, autosomal dominant polycystic kidney disease, neurofibromatosis type 1, Lehman syndrome, Ehlers-Danlos syndrome may play a role in the development of spontaneous intracranial hypotension [3] [5] [8]. Other possible causes of spinal CSF leak include bone spurs piercing the dura or iatrogenic causes [3].

Improved imaging modalities and increased awareness of clinical presentations among clinicians lead to more recognition of intracranial hypotension. Brain MRI, spinal MRI, noncontrast MR myelography and radioisotope cisternography are helpful to diagnose intracranial hypotension [1] [2] [4] [6] [7]. Common brain MRI findings in intracranial hypotension include diffuse thickening of the pachymeninges with Gadolinium enhancement, engorgement of venous sinuses, subdural fluid collections, enlargement of the pituitary gland, and downward displacement of the brain [1] [2] [3] [6] [7]. Monro–Kellie hypothesis stated that the sum of the volumes of intracranial blood, CSF, and brain

**Table 1.** Demographic and clinical characteristics of the patients.

| Case # | gender | age | Symptoms signs | CSF leak location | Epidural blood patch | Complicated with SDH |
|---|---|---|---|---|---|---|
| 1 | F | 33 | Orthostatic headache, dizziness | not detected | nil | No |
| 2 | F | 27 | Orthostatic headache, nausea, vomiting, photophobia | C1-2 | x1 | No |
| 3 | F | 61 | Orthostatic headache | T1-9 | x1 | No |
| 4 | M | 46 | Orthostatic headache, dizziness, nausea, vomiting | C2 | x2 | No |
| 5 | F | 46 | Orthostatic headache | T3-8 | x1 | No |
| 6 | F | 29 | Orthostatic headache, dizziness, tinnitus | C2 | x2 | No |
| 7 | M | 38 | Orthostatic headache, back pain, tinnitus | C-T-L spine | x2 | No |
| 8 | F | 41 | Orthostatic headache | mid T-spine | x1 | No |
| 9 | M | 59 | Orthostatic headache, dizziness | not detected | nil | No |
| 10 | M | 60 | Orthostatic headache | CSF leak was shown but the leakage site cannot be identified precisely | x1 | Yes |
| 11 | M | 21 | Orthostatic headache | CSF leak was shown but the leakage site cannot be identified precisely | nil | No |
| 12 | F | 30 | Orthostatic headache | CSF leak was shown but the leakage site cannot be identified precisely | x1 | No |
| 13 | M | 35 | Orthostatic headache, tinnitus | CSF leak was shown but the leakage site cannot be identified precisely | nil | No |
| 14 | F | 37 | Orthostatic headache, dizziness, nausea, vomiting | not detected | nil | No |
| 15 | F | 34 | Orthostatic headache | T12 to L3 | x1 | No |
| 16 | M | 63 | Orthostatic headache, neck stiffness | T4-8 | x1 | No |
| 17 | F | 40 | Orthostatic headache, vomiting | middle to lower T spine but the CSF leakage site cannot be identified precisely | x1 | Yes |
| 18 | F | 41 | Orthostatic headache, neck pain | not detected | x1 | No |
| 19 | F | 35 | Orthostatic headache, vertigo, vomiting | No spine MRI or MR myelography available | x1 | No |

**Continued**

| | | | | | | |
|---|---|---|---|---|---|---|
| 20 | F | 54 | Orthostatic headache, nausea, vomiting, phonophobia | L2-4 | x1 | No |
| 21 | F | 44 | Orthostatic headache | not detected | nil | No |
| 22 | M | 31 | Orthostatic headache, nausea, vomiting | not detected | nil | No |
| 23 | M | 30 | Orthostatic headache, dizziness | not detected | nil | No |
| 24 | F | 35 | Orthostatic headache, back pain | C1-2 | x2 | No |
| 25 | M | 44 | Orthostatic headache, nausea, vomiting, photophobia, neck stiffness | T5, L2 | x2 | Yes |
| 26 | M | 31 | Dizziness and vomiting, neck stiffness | C1-C2, T3 to T5 | x1 | No |
| 27 | M | 58 | Orthostatic headache, dizziness, neck stiffness, nausea | CSF leak was shown but the leakage site cannot be identified precisely | x1 | No |

tissue must remain constant in an intact cranium which can explain the brain MRI features [2] [3]. Spinal MRI findings in case of intracranial hypotension are extraarachnoid fluid collections, spinal meningeal enhancement, meningeal diverticula, dilation of the anterior internal vertebral (epidural) venous plexus [2] [6]. Noncontrast MR myelography using heavily T2-weighted three-dimensional sequences is performed without a dural puncture or intravenous gadolinium contrast and may be an effective technique to diagnose spontaneous intracranial hypotension. It may show extradural fluid collections or structural abnormality such as meningeal diverticula [6]. In addition, it does not carry the adverse risks of causing a CSF leak from dural puncture or potential adverse reactions to intrathecal gadolinium associated with myelography performed with intrathecal injection. Radioisotope cisternography is a useful procedure for identifying CSF leaks [2]. It may be performed when noninvasive MR myelography is nondiagnostic. A CSF leak can be detected either directly by accumulation of radioactivity outside the subarachnoid space or indirectly by a limited ascent of the tracer to the cerebral convexities and its early accumulation in the kidneys, urinary bladder, and soft tissues [2] [6].

The goal of treatment is to restore normal CSF pressure to achieve symptomatic relief [1]. Conservative treatment including bed-rest, fluid hydration, analgesics is initially provided [1]. Epidural blood patch, either targeted or non-targeted can stop CSF leak in most cases and is recommended when conservative treatment is not effective to achieve symptomatic relief [2] [4]. Under certain circumstances, surgical or endovascular closure of the CSF leak or fistula may be employed [2].

Chronic subdural hematoma is a common disorder in neurosurgery. Head

trauma, elderly, very young age, brain atrophy and bleeding tendency are risk factors to have chronic subdural hematoma. Clinically, when a patient without these risk factors presented with chronic subdural hematoma should remind the diagnosis of intracranial hypotension especially when the patient has postural headache [10]. The reported incidence of subdural hematoma among spontaneous intracranial hypotension ranges from 16% to 57% [9]. The pathogenesis may involve tearing of bridging veins or bleeding from enlarged veins in the subdural space because of low intracranial pressure and brain descent [8] [10]. Both epidural blood patch and surgical removal of the subdural hematoma are essential procedures to treat patients of intracranial hypotension associated with subdural hematoma especially when the subdural hematoma causes mass effect. However, which procedure should be done initially remains controversial [9].

Our case number is limited; all of these 27 patients had good clinical outcome after having either conservative treatment or epidural blood patch management. The imaging modalities of these 27 patients were brain MRI, spine MRI and MR myelography and none of them did radioisotope cisternography. The most frequent site of CSF leakage in this group of patient was T-spine followed by C-spine and then L-spine. Small case number and retrospectively review are the limitation of this study.

## 6. Conclusion

Back to this particular patient, we learn that recurrent CSF leakage from different sites can happen. Diffuse pachymeningeal enhancement detected on brain MRI in case of intracranial hypotension is reversible. Burr hole drainage of the massive subdural hematoma, epidural blood patch procedure, fluid hydration and bed rest are essential and effective to treat massive subdural hematoma associated with intracranial hypotension. Intracranial hypotension is a benign disease but its diagnosis requires high vigilance of the clinician.

## Conflicts of Interest

The author declares no conflicts of interest regarding the publication of this paper.

## References

[1] Rettenmaier, L.A., Park, B.J., Holland, M.T., Hamade, Y.J., Garg, S., Rastogi, R. and Reddy, C.G. (2017) Value of Targeted Epidural Blood Patch and Management of Subdural Hematoma in Spontaneous Intracranial Hypotension: Case Report and Review of the Literature. *World Neurosurgery*, **97**, 27-38. https://doi.org/10.1016/j.wneu.2016.09.076

[2] Paldino, M., Mogilner, A.Y. and Tenner, M.S. (2003) Intracranial Hypotension Syndrome: A Comprehensive Review. *Neurosurgical Focus*, **15**, 1-8. https://doi.org/10.3171/foc.2003.15.6.8

[3] Schmidt, M.H. and Binder, D.K. (2002) Spontaneous Intracranial Hypotension. *Contemporary Neurosurgery*, **24**, 1-4.

https://doi.org/10.1097/00029679-200209150-00001

[4] Dobrocky, T., Nicholson, P., Hani, L., Mordasini, P., Krings, T., Brinjikji, W., Cutsforth-Gregory, J.K., Schar, R., Schankin, C., Gralla, J., Pereira, V.M., Raabe, A., Farb, R., Beck, J. and Piechowiak, E.I. (2022) Spontaneous Intracranial Hypotension: Searching for the CSF Leak. *The Lancet Neurology*, **21**, 369-380. https://doi.org/10.1016/S1474-4422(21)00423-3

[5] Schievink, W.I. (2021) Spontaneous Intracranial Hypotension. *The New England Journal of Medicine*, **385**, 2173-2178. https://doi.org/10.1056/NEJMra2101561

[6] Schievink, W.I. (2006) Spontaneous Spinal Cerebrospinal Fluid Leaks and Intracranial Hypotension. *JAMA*, **295**, 2286-2296. https://doi.org/10.1001/jama.295.19.2286

[7] Wang, H.K., Liliang, P.C., Liang, C.L., Lu, K., Hung, K.C. and Chen, H.J. (2010) Delayed Subdural Hematoma after Epidural Blood Patching in a Patient with Spontaneous Intracranial Hypotension-Case Report. *Neurologia medico-chirurgica*, **50**, 479-481. https://doi.org/10.2176/nmc.50.479

[8] Kim, B.W., Jung, Y.J., Kim, M.S. and Choi, B.Y. (2011) Chronic Subdural Hematoma after Spontaneous Intracranial Hypotension: A Case Treated with Epidural Blood Patch on C1-2. *Journal of Korean Neurosurgical Society*, **50**, 274-276. https://doi.org/10.3340/jkns.2011.50.3.274

[9] Takahashi, K., Mima, T. and Akiba, Y. (2016) Chronic Subdural Hematoma Associated with Spontaneous Intracranial Hypotension: Therapeutic Strategies and Outcomes of 55 Cases. *Neurologia medico-chirurgica*, **56**, 69-76. https://doi.org/10.2176/nmc.oa.2015-0032

[10] Schievink, W.I., Maya, M.M., Moser, F.G. and Tourje, J. (2005) Spectrum of Subdural Fluid Collections in Spontaneous Intracranial Hypotension. *Journal of Neurosurgery*, **103**, 608-613. https://doi.org/10.3171/jns.2005.103.4.0608