Neuroradiology

Pontine bleeding following drainage of subdural hematoma in craniospinal hypotension

Temel Fatih Yılmaz MD[a,*], Ayse Aralasmak MD[b], Huseyin Toprak MD[b], Gulsen Kocaman MD[c], Alpay Alkan MD[b]

[a] Department of Radiology, Kars State Hospital, Yenişehir Mahallesi, İsmail Aytemiz Bulvarı, No:55, 36200 Kars, Turkey
[b] Department of Radiology, Bezmialem Vakif University, Istanbul, Turkey, Adnan Menderes Bulvarı, Vatan Caddesi, 34093 Fatih/İstanbul, Turkey
[c] Department of Neurology, Bezmialem Vakif University, Adnan Menderes Bulvarı, Vatan Caddesi, 34093 Fatih/İstanbul, Turkey

ARTICLE INFO

ABSTRACT

Craniospinal hypotension is the syndrome of orthostatic headache associated with low cerebrospinal fluid pressure. Imaging findings are usually explained by Monro-Kellie hypothesis stating that the craniospinal compartment is incompressible and any increase in volume of one of the craniospinal constituents (blood, cerebrospinal fluid, and parenchyma) must be compensated by a decrease in volume of another constituent or vice versa. We report a case of craniospinal hypotension in whom drainage of subdural hematoma upon clinical impairments resulted in pontine hemorrhage, supporting Monro-Kellie hypothesis.

Introduction

Craniospinal hypotension (CSH) is the syndrome of low pressure headache associated with low cerebrospinal fluid (CSF) pressure (<60 mm H_2O) in the absence of a known dural puncture. In case of dural puncture, it is called iatrogenic CSH. It is an uncommon syndrome known as Schaltenbrand syndrome or spontaneous aliquorrhea [1]. The great majority of patients are in the fifth or sixth decade of life, and the mean age is the early 40s [2]. The classic clinical hallmark of CSH is a postural or orthostatic headache that is exacerbated by standing and is relieved in the recumbent position. Patients may also present with diplopia, tinnitus, vertigo, dizziness, stiff neck, nausea, vomiting, hyperacusis, deafness, and visual loss [1,3]. Imaging studies, especially magnetic resonance imaging (MRI), are helpful in confirming the diagnosis and ruling out other entities. Imaging findings can be normal or include caudal displacement of cerebrum below tentorium, diffuse intense pachymeningeal enhancement, dural sinus distention, subdural effusion or
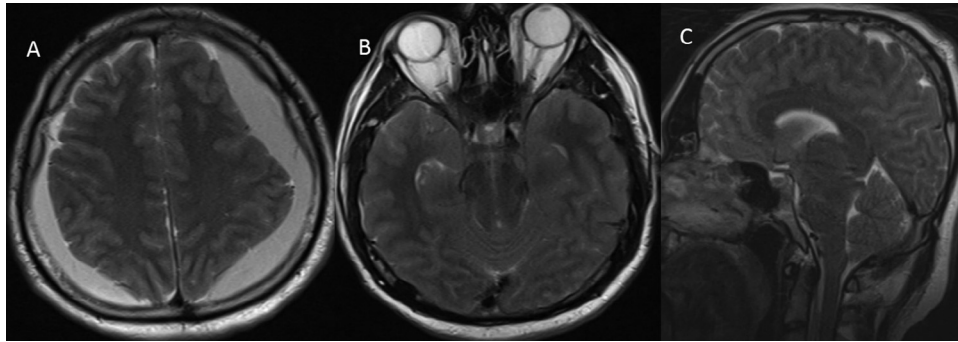
Fig. 1 – (A-C) Axial T2W (A) images show bilateral cerebral subdural effusion or hematoma. Axial (B) and saggital (C) T2W images demonstrate cerebellar tonsils at the level of foramen magnum with inferior displacement of brainstem and obliteration of perimesencephalic and suprasellar cistern and increase in anterior-posterior diameter of mesencephalon due to compression.

hematomas, cerebellar tonsillar herniation, enlarged edematous pituitary gland, obliteration of suprasellar and perimesencephalic cistern, dural sinus thrombosis due to long standing dural sinus distention, and spinal epidural congestion [1,4,5]. Imaging findings are easily explained by Monro-Kellie hypothesis, which states that the craniospinal and its constituents (blood, CSF, and parenchyma) create a state of volume equilibrium, such that any decrease in volume of one of the cranial constituents must be compensated by an increase in volume of another constituent or vice versa [6]. We are reporting a case of iatrogenic CSH in whom drainage of subdural hematoma upon clinical impairments before treatment of spinal leakage resulted in pontine hemorrhage, supporting Monro-Kellie hypothesis.

Case report

A 37-year-old man was admitted to our hospital with complaints of headache for 3 weeks, dizziness, and loss of orientation in short periods of time with no history of head trauma. His headache was aggravated by standing. He was hospitalized with a diagnosis of CSH. Cranial MRI revealed bilateral subdural effusions (Fig. 1). After a few days, his clinical situation became worse and his Glasgow Coma Scale score decreased. Follow-up brain MRI demonstrated increased subdural hematoma with fluid layering showing ongoing bleeding (Fig. 2). Because of decreasing Glasgow Coma Scale score and increasing subdural effusion, subdural hematomas were evacuated bilaterally with parietal approach, and hemorrhagic fluid was aspirated via burr hole. In postoperative period within 12 hours, the patient was confused and a newly developed left hemiparesis was added to his clinical situation. Diffusion MRI and unenhanced cranial computed tomography scan were performed. There was pontine hemorrhage (Fig. 3). Routine cervical and thoracic spinal MRI was performed. Epidural congestion was noted (Fig. 4). MR cisternography was performed through L4-L5 interspinous space. The contrast material was injected into subarachnoid distance after observing CSF flow coming out of the needle. The opening CSF pressure was low (50 mm H$_2$O). MR cisternography demonstrated intense epidural contrast extravasation from subarachnoid

space throughout cervical and upper thoracic level (from C3 to T8) (Fig. 5). Epidural leakage was probably secondary to dural tear. On pre- and postinjection MRI, there was no epidural congestion nor subdural or epidural contrast extravasation in lumbal region that was the contrast injection site for MR cisternography. Epidural blood patch was performed 3 times. At the end of the 3 sessions, significant improvement of the patient's clinical situation with resolution of spinal epidural leakage and reduction in brain subdural effusions was noted.

Discussion

CSH is an underrecognized cause of headache, resulting from persistent CSF leak. Diagnosis hinges on the history of orthostatic headache, low CSF opening pressure, and characteristic
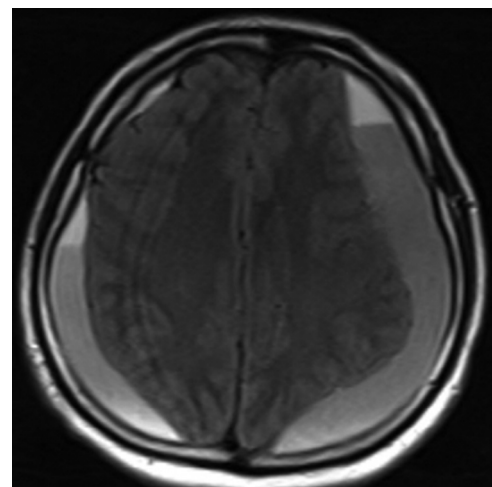

Fig. 2 – On control cranial MRI performed in a few days because of clinical worsening and decrease in Glasgow Coma Scale, axial T2W images show an increase in subdural hematomas with fluid-fluid levels due to ongoing bleeding. Subdural hematoma was drained to relieve pressure on brainstem.
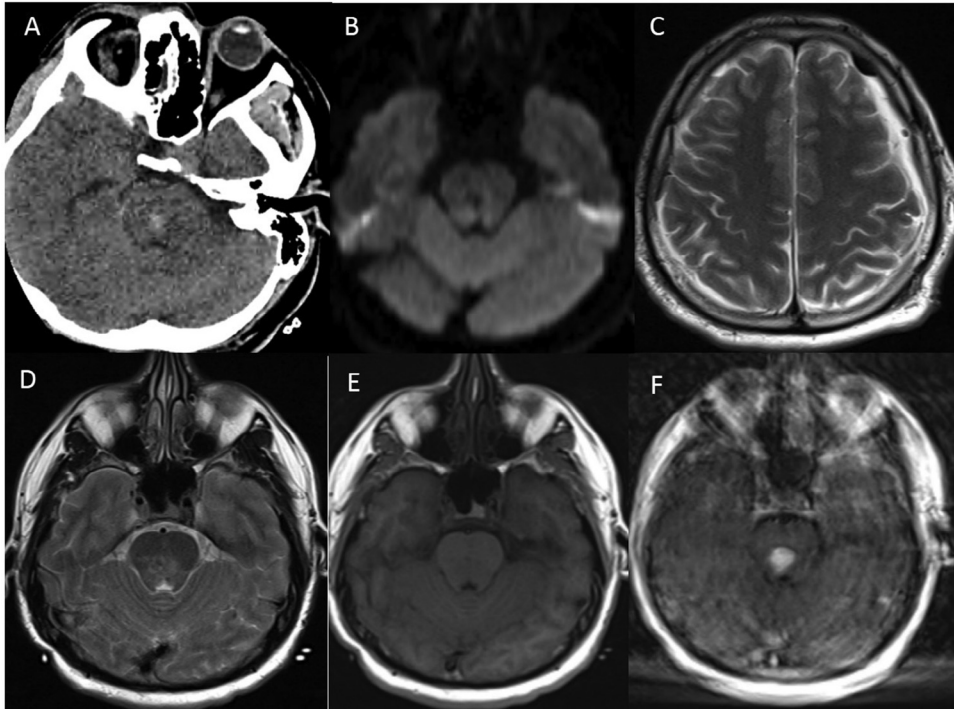
Fig. 3 – (A-F) After subdural drainage, confusion and left hemiparesis developed. On cranial CT (A) and DWI (B), pontine hemorrhage was detected. Axial T2W (C) images show reductions in bilateral cerebral subdural effusions. Axial T2W (D) and pre (E) and postcontrast T1W (F) images show pontine edema with intensive contrast enhancement by virtue of extravasation.

Fig. 4 – (A-D) On saggital T2W (A, C) and T1W (B, D) cervical and thoracic MRI, 2 different fluid intensity is seen around the spinal cord at the cervical and thoracic regions. Epidural space (long arrows) appears brighter than subarachnoid space (dashed long arrows) on T2W. Stagnant fluid with no flow inside is the cause of this brightness in epidural place, comparing the free flowing CSF in subarachnoid space. Epidural fat in the thoracic region (short arrows) seems hyperintense on T1 (D) and hypointense on T2(C), comparing the epidural fluid accumulation. Epidural fluid accumulation throughout cervical and thoracic regions is a sign of epidural leakage or congestion.

Fig. 5 – (A-F) After intrathecal gadolinium injections, fat-saturated T1W views of MR cisternography demonstrated contrast leakage from subarachnoid region into epidural space throughout cervical and upper thoracic regions from C3 to T8 level (A, B). Arrows show contrast within epidural place and dashed arrows show normal contrast within subarachnoid level (A, B). Gadolinium in the epidural region is more intense from that in subarachnoid space (arrows on A-F). Subarachnoid space in lower thoracic and lumbar region appear flow void due to CSF flow (dashed arrows on C-F). Epidural leakage throughout lower cervical and upper thoracic spine is probably due to dural tear. There is no epidural leakage in the lumbar region, although intrathecal gadolinium was injected with a lumbar puncture at L4-L5 level (not seen here). Epidural blood patch was performed three times. On follow-up, the patient was discharged with marked clinical improvement.

brain MRI findings [1,4]. The etiology of CSH could not be determined easily but it is thought to be caused by probable weakness of spinal membranes. Dural leak can result from a disc herniation, a cervical bone spur, a minor head trauma, vigorous exercise, sexual activity, or a violent bout of coughing or sneezing, meningeal diverticula, tears in nerve root sleeves, or Tarlov cysts, and sometimes it is associated with an underlying connective tissue disorder [1,4].

In CSH, the role of lumber punction to demonstrate low CSF pressure and the timing of further investigations to identify a leak are recently debated because they require a dural puncture that may worsen the patient's condition. Diagnosis is usually made by clinical history and characteristic MRI findings [1,4,5]. Spinal findings in CSH are rare, can be normal, or include epidural congestion, narrowed and festooned thecal sac, or crowded appearance of cauda equina fibers in lumbosacral region [1,7,8]. However, spinal epidural congestion is not specific; it may also be seen in spinal muscular atrophy as in Hirayama disease, in connective tissue disorders as in Marfan disease, and in vascular pathologies as in abdominal inferior vena cava thrombosis or occlusion [7,9,10]. In CSH, spinal epidural congestion on MRI can be the result of low CSF pressure or CSF leak. For differentiation, MR cisternography with intrathecal gadolinium or radioisotope myelocisternography can be

performed. For the fine detail for localization of spinal leakage for surgical approach, CT cisternography with nonionic iodinated contrast material has better localizing power than radioisotope myelocisternography and MR cisternography [8].

The Monro-Kellie hypothesis explains both the spinal and the cranial dural congestion in CSH. The pressure-volume relationship between intracranial pressure, volume of CSF, blood, and parenchyma and craniospinal perfusion pressure is known as the Monro-Kellie hypothesis. The Monro-Kellie hypothesis states that the craniospinal compartment is incompressible, and the volume inside the craniospinal region is fix. The craniospinal and its constituents (blood, CSF, and parenchyma) create a state of volume equilibrium, such that any increase in volume of one of the craniospinal constituents must be compensated by a decrease in volume of another constituent or vice versa [6]. So, reduced intracranial pressure results in increased cerebral blood volume, which is strongly the cause of subdural hematoma seen in patients with CSH. In our case, abrupt clinical deterioration and increase subdural hematoma with active bleeding inside misdirect us to neurosurgeons for evacuation of subdural hematomas. Our patient's deterioration was due to aggravation of CSH, but we thought that his deterioration was due to

mass effect of subdural hematomas compressing the brainstem. Upon this thought, the patient was operated on. Following evacuation of subdural hematomas, the patient became worse since pontine bleeding occurred. Spinal MRI revealed spinal epidural congestion. Spinal leakage was searched with MR cisternography. Cervical and upper thoracic CSF leakage was found. Pontine bleeding in our case is also explained by Monro-Kellie hypothesis. Pontine bleeding was due to unbalanced intradural negative pressure that resulted in extravasation secondary to drainage of subdural hematomas without closure of spinal epidural CSF leakage. Extravasation like subdural effusion or pontine hemorrhage is to compensate intracranial hypotension by increasing cerebral blood volume according to Monro-Kellie hypothesis. Pontine hemorrhage can be a result of hypertension, amyloid angiopathy, vascular malformations, and anticoagulative treatment. None of them was present in our case but craniospinal hypotension. Pontine hemorrhage in CSH can be explained by Monro-Kellie hypothesis and has never been reported during the course of the CSH.

In the treatment of CSH, bed rest, increased fluid, and caffeine intake are suggested. If these choices are ineffective, spinal epidural blood patch should be performed. Symptoms caused by thecal compression and increased CSF pressure following epidural blood patch improve quickly.

As a conclusion, orthostatic headache is a major clinical symptom of CSH. The main MRI findings are diffuse intense pachymeningeal enhancement, dural sinus distention, subdural effusion or hematomas, cerebellar tonsillar herniation, and spinal epidural congestion. Evacuations of subdural hematomas in CSH without correction of spinal dural leakage can result in pontine bleeding. This finding is compatible with Monro-Kellie doctrine.

REFERENCES

[1] Haritanti A, Karacostas D, Drevelengas A, Kanellopoulos V, Paraskevopoulou E, Lefkopoulos A, et al. Spontaneous intracranial hypotension: clinical and neuroimaging findings in six cases with literature review. Eur J Radiol 2009;69:253–9.

[2] Schievink WI, Maya MM, Louy C, Moser FG, Sloninsky L. Spontaneous intracranial hypotension in childhood and adolescence. J Pediatr 2013;163:504–10.

[3] Forghani R, Farb RI. Diagnosis and temporal evolution of signs of intracranial hypotension on MRI of the brain. Neuroradiology 2008;50:1025–34.

[4] Fullam L, Saidha S, Harrington H, MacEneaney P. Spontaneous intracranial hypotension. Ir J Med Sci 2011;180:587–8.

[5] Allmendinger AM, Lee TC. Spontaneous intracranial hypotension from calcified thoracic disc protrusions causing CSF leak successfully treated with targeted epidural blood patch. Clin Imaging 2013;37:756–61.

[6] Smith ER, Madsen JR. Cerebral pathophysiology and critical care neurology: basic hemodynamic principles, cerebral perfusion, and intracranial pressure. Semin Pediatr Neurol 2004;11:89–104.

[7] Dietemann JL, Bogorin A, Abu Eid M, Sanda R, Mourao Soares I, Draghici S, et al. Tips and traps in neurological imaging: imaging the perimedullary spaces. Diagn Interv Imaging 2012;93:985–92.

[8] Chiapparini L, Ciceri E, Nappini S, Castellani MR, Mea E, Bussone G, et al. Headache and intracranial hypotension: neuroradiological findings. Neurol Sci 2004;25(Suppl. 3):S138–41, Review.

[9] Gandhi D, Goyal M, Bourque PR, Jain R. Case 68: Hirayama disease. Radiology 2004;230:692–6.

[10] Chun JY, Dillon WP, Berger MS. Symptomatic enlarged cervical anterior epidural venous plexus in a patient with Marfan syndrome. AJNR Am J Neuroradiol 2002;23:622–4.