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A small leak will sink the brain: targeted C1-C2 patching

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Abstract

Background

Spontaneous intracranial hypotension (SIH) syndrome results from spontaneous spinal CSF leaks. The first treatment of choice consists of lumbar epidural blood patching. If this fails further imaging is mandatory to explore the possibility of targeted therapy.

Case Description

We describe a case of a 50-year old woman who developed spontaneous intracranial hypotension after minor blunt cervical trauma, complicated with bilateral subdural hematomas. Two lumbar epidural blood patches were unsuccessful. MRI with intrathecal gadolinium revealed a CSF leak at the C1-C2 level. A targeted blood patch via a percutaneous high thoracic epidural approach was performed and symptoms disappeared in the immediate postoperative period with a regression of the subdural hematomas on subsequent imaging.

Conclusions

A targeted epidural blood patch using an epidural catheter represents an elegant approach to a CSF leak at the C1-C2 region and can be successful in treating patients with severe intracranial hypotension syndrome.

Introduction

Spontaneous intracranial hypotension (SIH) can occur after trivial trauma but in many cases no clear eliciting factor is present. SIH syndrome typically results from spontaneous spinal CSF leaks of which three types have been proposed: the dural tear, the meningeal diverticulum and the CSF-venous fistula.¹ The precise cause of these CSF leaks is unknown, but an underlying structural weakness of the spinal meninges is suggested.² A recent study also stresses the importance of degenerative changes and calcifications which can damage a normal dura.³ The diagnosis of SIH is made after a typical clinical history of orthostatic headache with or without associated symptoms, imaging showing typical MRI signs and sometimes lumbar puncture with measurement of a low opening pressure.⁴

In many cases, symptoms are self-limiting and can be treated with conservative measures such as bed rest, analgesics, hydration and caffeine intake. If symptoms do not resolve with conservative measures, a lumbar epidural blood patch will be successful in many patients. When symptoms persist, a second lumbar epidural blood patch with the injection of a higher volume of blood can be considered. In patients with unsuccessful lumbar blood patching further imaging to explore the possibility of targeted treatment is mandatory.

We present the successful treatment of a posttraumatic CSF leak at the C1-C2 level using a targeted epidural blood patch through an Ascenda catheter. Imaging findings as well as treatment options are discussed.

Case

After sustaining a minor cervical trauma, by walking into a blunt wooden object, a 50-year old woman developed severe occipital headache, associated with nausea and vomiting. Initially, she reported tinnitus and orthostatic headache, followed by a more continuous headache. MR imaging of the brain showed bilateral subdural hematomas (Figure 1A), diffuse meningeal thickening and brain sagging, suggesting intracranial hypotension. Two lumbar epidural blood patches were unsuccessful. Symptoms persisted for four months after which the patient was referred to our center.

High resolution MR imaging of the spine did not reveal any abnormalities. MRI with intrathecal gadolinium, T1-weighted imaging with fat suppression acquired one hour after injection of 0.5 ml gadolinium (Dotarem, Guerbet), revealed a CSF leak through a bilateral meningeal defect at the C1-C2 level more prominent at the left side (Figure 2). A targeted treatment was initiated consisting of a blood patch via a percutaneous high thoracic epidural approach using the Ascenda (Medtronic, Ø 1.2 mm) catheter. The awake patient was put in a prone position and the catheter was guided upwards to the C2 level (Figure 1C) after which 10 ml of autologous venous blood was injected in the left epidural space. During injection the patient was carefully monitored for potential signs of spinal cord compression. All symptoms disappeared in the immediate postoperative period with a regression of the subdural hematomas on subsequent imaging. Six months after the procedure the patient remained pain free and MRI of the brain (Figure 1B) showed no abnormalities.

Discussion

Intrathecal contrast enhanced imaging can be indicated to reliably detect a CSF leak and target treatment in SIH syndrome. Computed tomography (CT), magnetic resonance (MR) or digital subtraction myelography are the most commonly applied imaging modalities in this respect.⁵ For fast CSF leaks, CT and MR myelography are less sensitive and a modality with higher temporal resolution should be used which comprise dynamic CT myelography and Digital subtraction myelography (DSM). DSM remains the golden standard for detecting CSF leaks, because of its high spatial and temporal resolution.^{6, 7} In our case, we used MR myelography with intrathecal gadolinium (0.5 ml, Dotarem, Guerbet) revealing a leak at the C1-C2 level. The use of intrathecal gadolinium is off-label; no adverse effects however have been reported with the use of small doses.^{8, 9} Leaks at the C1-C2 level have been reported in the literature¹⁰⁻¹⁵, probably due to mechanical factors associated with head rotation. One must be aware of the potential false localizing sign when visualizing contrast at the C1-C2 level, which may be the result of a more caudal CSF leak.^{16, 17} The C1-C2 level may be more vulnerable for accumulation of CSF due to the anatomical differences at this level related to head rotation. We believe it is important to not only visualize the contrast but also to identify the dural defect on imaging.

Once the exact location of the leak is identified, targeted treatment can be initiated, either consisting of neurosurgical repair or a targeted epidural blood patch. We believe a targeted epidural blood patch represents the first choice of treatment because of the lower inherent risks involved. Even high leaks in the cervical spine can be accessed with an epidural catheter.^{10, 15} It is moreover important to note that complex surgical anatomy of some dural defects can prohibit primary closure during surgery.¹⁸ In refractory cases, surgery can be necessary to address the leak site, especially if underlying calcifications ('microspurs') are present. In these cases the CSF leak is unlikely to recover if the causative lesion is not removed.³

Conclusion

A targeted epidural blood patch using an epidural catheter represents a less invasive approach to a CSF leak at the C1-C2 region and can be successful in treating patients with severe intracranial hypotension syndrome.

Figure legend

Figure 1. *A.* Preoperative T2-weighted MR imaging showing bilateral subdural hematomas. *B.* Complete resolution of subdural hematomas on T2-weighted imaging 6 months after targeted epidural blood patch. *C.* Intraoperative fluoroscopic AP view of the catheter tip (arrow) at the C2 level.

Figure 2. *A.* Sagittal, *B.* Coronal, *C.* Axial MRI of the spine, T1-weighted imaging with fat suppression, acquired one hour after intrathecal injection of gadolinium shows the CSF leak at C1-C2 level. A dural defect is seen on panel C.

References

1. Schievink WI, Maya MM, Jean-Pierre S, Nuno M, Prasad RS, Moser FG. A classification system of spontaneous spinal CSF leaks. *Neurology* 2016;87:673-679.
2. Schievink WI, Meyer FB, Atkinson JL, Mokri B. Spontaneous spinal cerebrospinal fluid leaks and intracranial hypotension. *Journal of neurosurgery* 1996;84:598-605.
3. Beck J, Ulrich CT, Fung C, et al. Diskogenic microspurs as a major cause of intractable spontaneous intracranial hypotension. *Neurology* 2016;87:1220-1226.
4. Schievink WI. Spontaneous spinal cerebrospinal fluid leaks and intracranial hypotension. *Jama* 2006;295:2286-2296.
5. Kranz PG, Luetmer PH, Diehn FE, Amrhein TJ, Tanpitukpongse TP, Gray L. Myelographic Techniques for the Detection of Spinal CSF Leaks in Spontaneous Intracranial Hypotension. *AJR American journal of roentgenology* 2016;206:8-19.
6. Hoxworth JM, Patel AC, Bosch EP, Nelson KD. Localization of a rapid CSF leak with digital subtraction myelography. *AJNR American journal of neuroradiology* 2009;30:516-519.
7. Hoxworth JM, Trentman TL, Kotsenas AL, Thielen KR, Nelson KD, Dodick DW. The role of digital subtraction myelography in the diagnosis and localization of spontaneous spinal CSF leaks. *AJR American journal of roentgenology* 2012;199:649-653.
8. Albayram S, Kilic F, Ozer H, Baghaki S, Kocer N, Islak C. Gadolinium-enhanced MR cisternography to evaluate dural leaks in intracranial hypotension syndrome. *AJNR American journal of neuroradiology* 2008;29:116-121.
9. Chazen JL, Talbott JF, Lantos JE, Dillon WP. MR myelography for identification of spinal CSF leak in spontaneous intracranial hypotension. *AJNR American journal of neuroradiology* 2014;35:2007-2012.
10. Kwon SY, Kim YS, Han SM. Spontaneous C1-2 cerebrospinal fluid leak treated with a targeted cervical epidural blood patch using a cervical epidural Racz catheter. *Pain physician* 2014;17:E381-384.
11. Sykes KT, Yi X. Intracranial hypotension headache caused by a massive cerebrospinal fluid leak successfully treated with a targeted c2 epidural blood patch: a case report. *Pain physician* 2013;16:399-404.
12. Rai A, Rosen C, Carpenter J, Miele V. Epidural blood patch at C2: diagnosis and treatment of spontaneous intracranial hypotension. *AJNR American journal of neuroradiology* 2005;26:2663-2666.
13. Smith KA. Spontaneous intracranial hypotension: Targeted or blind blood patch. *Journal of clinical neuroscience : official journal of the Neurosurgical Society of Australasia* 2016;25:10-12.
14. Falatko SR, Kelkar P, Setty P, Tong D, Soo TM. C1-C2 cryptic cerebrospinal fluid leak directly identified by pressurized radionuclide cisternography: Case report and review of the literature. *Surgical neurology international* 2015;6:126.
15. Wang E, Wang D. Successful treatment of spontaneous intracranial hypotension due to prominent cervical cerebrospinal fluid leak with cervical epidural blood patch. *Pain medicine* 2015;16:1013-1018.
16. Yousry I, Forderreuther S, Moriggl B, et al. Cervical MR imaging in postural headache: MR signs and pathophysiological implications. *AJNR American journal of neuroradiology* 2001;22:1239-1250.
17. Schievink WI, Maya MM, Tourje J. False localizing sign of C1-2 cerebrospinal fluid leak in spontaneous intracranial hypotension. *Journal of neurosurgery* 2004;100:639-644.
18. Cohen-Gadol AA, Mokri B, Piepgras DG, Meyer FB, Atkinson JL. Surgical anatomy of dural defects in spontaneous spinal cerebrospinal fluid leaks. *Neurosurgery* 2006;58:ONS-238-245; discussion ONS-245.

Highlights:

- Case of severe spontaneous intracranial hypotension with subdural hematomas
- Lumbar blood patching was unsuccessful
- MR myelography with intrathecal gadolinium revealed a CSF leak at the C1-C2 level
- A targeted blood patch using an epidural catheter was performed
- Resolution of symptoms within days, 6m later still pain free with normal head MRI

Abbreviations List:

CSF	cerebrospinal fluid
CT	computed tomography
DSM	digital subtraction myelography
MR	magnetic resonance
MRI	magnetic resonance imaging
SIH	spontaneous intracranial hypotension