Doi: 10.5152/TJAR.2021.676

Turkish Journal of Anaesthesiology & Reanimation

Intracranial Hypotension, Subdural Haematoma and Sjogren Syndrome

Manbir Kaur, Priyanka Sethi, Pradeep Bhatia
Department of Anesthesia & Critical Care, All India Institute of Medical Sciences (AIIMS), Jodhpur, India

Cite this article as: Kaur M, Sethi P, Bhatia P. Intracranial Hypotension, Subdural Hematoma and Sjogren Syndrome. *Turk J Anaesthesiol Reanim.* 2021;49(4):344-345.

Dear Editor,

We read with great interest the article by Yildirim et al.[1] published recently in the *Turkish Journal of Anesthesiology & Reanimation*. The authors report an extremely rare case of a 61-year-old female patient of Sjogren syndrome (SS) having postdural puncture headache and acute subdural hematoma (SDH) following the spinal anaesthesia. The spinal anaesthesia was given for performing cystoscopy as the patient also suffered from bladder cancer. The authors are to be praised for keen observation of all these events in a patient having connective tissue disorder and then trying to explain the link between them. We agree with the authors on many points, but wish to add and clarify some of their observations.

First, the subdural haematoma appears to be chronic rather than acute. It is very thin as per the images provided. There is no evidence of any mass effect. It looks more of the subdural hygroma or chronic SDH rather than acute haematoma due to its density (as acute SDH by definition is hyperdense on computed tomography [CT] scan). Such a minimal blood collection with no mass effect is unlikely to present with a severe headache or by the change in the character of the headache, as reported by the authors. This minimal collection could have also been present before the spinal anaesthesia as such a minimal, latent subdural haematoma is not uncommon at this age.[2] Therefore, in addition to SDH, other possibilities like migraine, subarachnoid haemorrhage, meningitis and cerebral venous thrombosis should also be kept in the differentials of headache in this setting.[3] That is why, in addition to radiological investigations, the serial neurological examination should always be stressed.[4]

Second, the authors placed an epidural blood patch despite the patient having relief in her postural headache. She did not have any severe persistent headaches or any other focal neurological deficits. They did it prophylactically to decrease the risk of further intracranial bleeding owing probably to persistent spinal cerebrospinal fluid (CSF) leakage. However, is it warranted in every such case that remains to be proven? Some authors argue that the epidural blood patch performed in such cases could lead to rebound intracranial hypertension and neurological deficits.[5–7] Such a small haematoma could, therefore, have also been managed conservatively without the application of a blood patch. It could be thought of as analogous to unnecessary spinal procedures being done these days.[8] Furthermore, the second brain CT image cut showing resolution of hematoma (after 4 days of the epidural patch) is not taken at the same level as the first CT image.

Third, the authors speculate that the connective tissue disturbance in the dura of SS patients is the primary factor in the formation of SDH. However, studies have shown that the age-related brain atrophy and the brain sagging due to decreased CSF volume and pressure caused by spinal CSF leak are the significant factors in the pathophysiology of SDH in this population.[3,9] Both these factors (present in the reported patient) will increase the gap between the brain surface and the dura, causing increased stretching and consequent rupture of the bridging veins. However, we agree with the authors that the connective tissue defect in SS may lead to delayed healing of the spinal dural defect resulting in persistent CSF leak, thus contributing to the formation of SDH.

Furthermore, last but not least, authors have linked the idiopathic hypertrophic pachymeningitis, intracranial hypotension and the SS. They propose that the inflammation of dura in their patient due to idiopathic

Corresponding Author: Manbir Kaur E-mail: doctor.manbir@gmail.com

Received: May 7, 2020 Accepted: June 12, 2020
Available Online Date: August 3, 2021

hypertrophic pachymeningitis might have led to delayed dural defect closure causing persistent CSF leak and consequent SDH. However, idiopathic hypertrophic pachymeningitis is altogether a different entity and has no relation with intracranial hypotension due to spinal CSF leak.[10] The dural enhancement in intracranial hypotension due to spinal CSF leak has two cardinal features: It is smooth and diffuse. In contrast to this, the dural enhancement in idiopathic hypertrophic pachymeningitis is typically irregular and does not involve the dura uniformly.[10] Furthermore, since we do not have the magnetic resonance images as proof for the presence of hypertrophic pachymeningitis, it is presumptive to propose its occurrence in this patient.

Interestingly, the inflammatory response of the dura in SS is the exact opposite of another rare connective tissue disorder, the Langerhans cell histiocytosis (LCH), whereas in LCH, there is excessive proliferation of the granulation tissue in the dura, the dura presumably does not heal in the SS due probably to lack of formation of acceptable granulation tissue.[11,12]

Peer-review: Externally peer-reviewed.

Author Contribution: Concept - M.K.; Design - M.K.; Supervision - P.S.; Interpretation - P.D.; Literature Search - P.S.; Writing - M.K.; Critical Reviews - P.B.

Conflicts of Interest: The authors have no conflicts of interest to declare.

Financial Disclosure: The authors declared that this study has received no financial support.

References

1. Yıldırım HU, Bakır M, Atıcı ŞR. Postdural puncture headache and acute subdural haematoma in Sjögren's syndrome. *Turk J Anaesthesiol Reanim*. 2020;48(1):75-78. [CrossRef]
2. Yang W, Huang J. Chronic subdural hematoma: Epidemiology and natural history. *Neurosurg Clin N Am*. 2017;28(2):205-210. [CrossRef]
3. Kale A, Emmez H, Pişkin Ö, et al. Postdural puncture subdural hematoma or postdural puncture headache?: Two cases report. *Korean J Anesthesiol*. 2015;68(5):509-512. [CrossRef]
4. Bhaskar S, Gosal JS, Garg M, et al. Letter: The neurological examination. *Oper Neurosurg (Hagerstown)*. 2020; pii:opaa047. [CrossRef]
5. Rudehill A, Gordon E, Rahn T. Subdural haematoma. A rare but life-threatening complication after spinal anaesthesia. *Acta Anaesthesiol Scand*. 1983;27:376-377. [CrossRef]
6. Kardash K, Morrow F, Beique F. Seizures after epidural blood patch with undiagnosed subdural hematoma. *Reg Anesth Pain Med*. 2002;27:433-436. [CrossRef]
7. Zeidan A, Farhat O, Maaliki H, et al. Does postdural puncture headache left untreated lead to subdural hematoma? Case report and review of the literature. *Int J Obstet Anesth*. 2006;15(1):50-58. [CrossRef]
8. Bhaskar S, Gosal JS, Garg M, et al. Letter to the editor regarding "why would two patients with no disease be offered unnecessary transforaminal lumbar interbody fusions?" *Surg Neurol Int*. 2019;10:238. [CrossRef]
9. Gosal JS, Gurmey T, Kursa GK, et al. Is acetazolamide really useful in the management of traumatic cerebrospinal fluid rhinorrhea? *Neurol India*. 2015;63(2):197-201. [CrossRef]
10. Kranz PG, Gray L, Amrhein TJ. Spontaneous intracranial hypotension: 10 myths and misperceptions. *Headache*. 2018;58(7):948-959. [CrossRef]
11. Gosal JS, Ruparelia J, Garg M, et al. Giant skull vault defect in a child caused by Langerhans cell histiocytosis: The "calvaria-eating disease." *Childs Nerv Syst*. 2020;36(3):651-654. [CrossRef]
12. Gosal JS, Garg M, Bhaskar S, et al. Letter to the editor regarding "Langerhans cell histiocytosis of the skull in a burr hole site covered with hydroxyapatite material". *World Neurosurg*. 2020;134:667. [CrossRef]