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Case Report

A rare case of iatrogenic intracranial hypotension due to a minor CSF leakage ☆,☆☆

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ABSTRACT

Intracranial hypotension (IH) represents a rare complication, mainly following cerebrospinal fluid (CSF) leakage at the thoracic or cervicothoracic junction level. Iatrogenic IH may be expected secondary to the previous surgery or other procedures invading the patient's dura. Magnetic resonance imaging (MRI), computerized tomography (CT) scan images, CT cisternography, and magnetic resonance cerebrospinal fluid flow (MR CSF) remains the modality of choice to establish the diagnosis. The patient is in her late sixth decade, reflecting a history of progressive headaches, nausea, and vomiting. Once a diagnosis of foramen magnum meningioma was established using MRI, total microscopic resection was applied. Brain sagging and subdural fluid collection were identified on postoperative day three, suggesting intracranial hypotension due to cerebrospinal fluid leakage. Diagnosing IH following the CSF leak during the postoperative phase remains challenging. Although rare, early clinical suspicion must be considered to establish the diagnosis.

Abbreviations: IH, intracranial hypotension; CSF, cerebrospinal fluid; MRI, magnetic resonance imaging; CT, computerized tomography scan; MR CSF, Magnetic resonance Cerebrospinal fluid flow.

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Introduction

Intracranial hypotension (IH) is a rare essential complication mainly due to cerebrospinal fluid (CSF) leakage at the thoracic or cervicothoracic junction level. This condition has an estimated incidence of 5 per 10,000 per year [1]. IH can be primary (spontaneous) or secondary (iatrogenic or traumatic). Iatrogenic IH can occur following the previous surgery or other procedures that invade the dura, such as the lumbar puncture or drain placement [1,2]. It is estimated that 16% of CSF leakages originated from recent surgical procedures [3]. Following a decrease in cerebrospinal fluid volume and intracranial pressure, complications such as subdural hematoma, subdural fluid collection, pituitary hyperemia, downward displacement of the brain hemispheres, and sagging of the cerebellum and brainstem may occur. These complications are distinguishable on magnetic resonance imaging (MRI) and computerized tomography (CT) scan images.

Furthermore, CT cisternography or MR CSF flow studies could assist us in detecting the leakage site [4]. These patients may be managed conservatively using bed rest, decreased activity, and over-hydration. In the case of iatrogenic or refractory IH, CT-guided injection of fibrin glue to the site of CSF leak or open surgery aiming for leakage site closure would be the best treatment option [5–7]. Here, the author reports a rare case of iatrogenic intracranial hypotension subsequent to previous surgery and discusses related literature.

Case presentation

In July 2019, a 58-year-old woman presented with progressive headache, nausea, and vomiting from a month ago. Her general condition and physical examination were uneventful. The patient underwent a brain MRI with and without gadolinium, revealing evidence of a foramen magnum meningioma. A few days later, she underwent surgery via suboccipital craniotomy, and under the guidance of neuromonitoring, total microscopic resection of meningioma was performed. Her general condition significantly improved after the operation, and no evidence of hematoma or surgical site collection was detected.

On the third postoperative day, she developed a headache aggravated in the upright position, swallowing difficulties, and gait disturbance. The patient was re-evaluated by a CT scan and MRI, which revealed evidence of subdural fluid collection. The before-mentioned findings favor intracranial hypotension due to cerebrospinal fluid leakage (Fig. 1).

Despite the lack of clear evidence of a CSF Leak, the patient underwent a brain CSF flow study MRI to detect any probable dural defect. CSF flow study revealed a 2.5 mm defect in the dura of the craniocervical junction adjacent to the posterior aspect of the tip of the right cerebellar tonsil, and apparent CSF flow through the defect was detected (Fig. 2). Even though there was no proper concordance between the patient's symptoms and a small source of CSF leak, according to the before-mentioned findings, the patient underwent surgery. Duraplasty was performed, and after the surgery, all symptoms improved significantly. The patient was discharged in stable condition after several days. In a 2-year follow-up, there was no evidence of intracranial hypotension or recurrence of the mass in the clinical examination and MRI (Fig. 3).

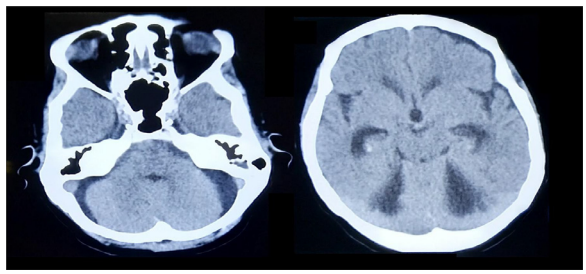

Fig. 1 – Axial noncontrast postoperative Brain CT demonstrates supratentorial and infratentorial subdural fluid collection, suggesting intracranial hypotension.

Discussion

Accurate diagnosis of CSF leak and intracranial hypotension in the postoperative stage is challenging and may be delayed or missed. Symptoms are often misleading, and there is no clear evidence of hypotension or leakage in some cases. Imaging evidence of intracranial hypotension will guide us to the diagnosis. However, these findings may not occur in the early stages or may be inconclusive. The most common symptom of intracranial hypotension is orthostatic headache, which only occurs in 23% of cases [8].

On the other hand, aggravation of headache in an upright position is more common, while the site of CSF leakage is in the spinal dura. Accordingly, orthostatic headache is not a differentiating sign in the diagnosis following cranial surgery. Other symptoms, such as vertigo, nausea, vomiting, dizziness, and loss of consciousness, are unspecific in the clinical course, and this makes the diagnosis more difficult, especially in the postoperative stages, when the patient may not be at a good level of awareness [9].

Imaging findings, including brain sagging, patchy meningeal enhancement, subdural hematoma or collection, tonsillar ectopia, shortened pontomamillary distance, elongated anteroposterior midbrain diameter, and pituitary hyperemia have a paramount role in the diagnosis (Fig. 4) [10]. Measurement of the CSF pressure is misleading since 34% of confirmed CSF leak cases have CSF pressure lower than 6 cm H_2O [11]. CSF leak should be counted and evaluated as a crucial differential diagnosis in postoperative intracranial hypotension.

The actual location of the leakage could be either in the cranial dura/surgical site or in the spinal dura following a lumbar puncture or other predisposing factors [12]. Locating the area of leakage could be challenging as well. MR CSF flow studies, CT cisternography, or radioisotope cisternography would be helpful in such circumstances. CT cisternography is more sensitive than MR CSF flow study and radioisotope cisternography. Besides, it would be more beneficial in determining the

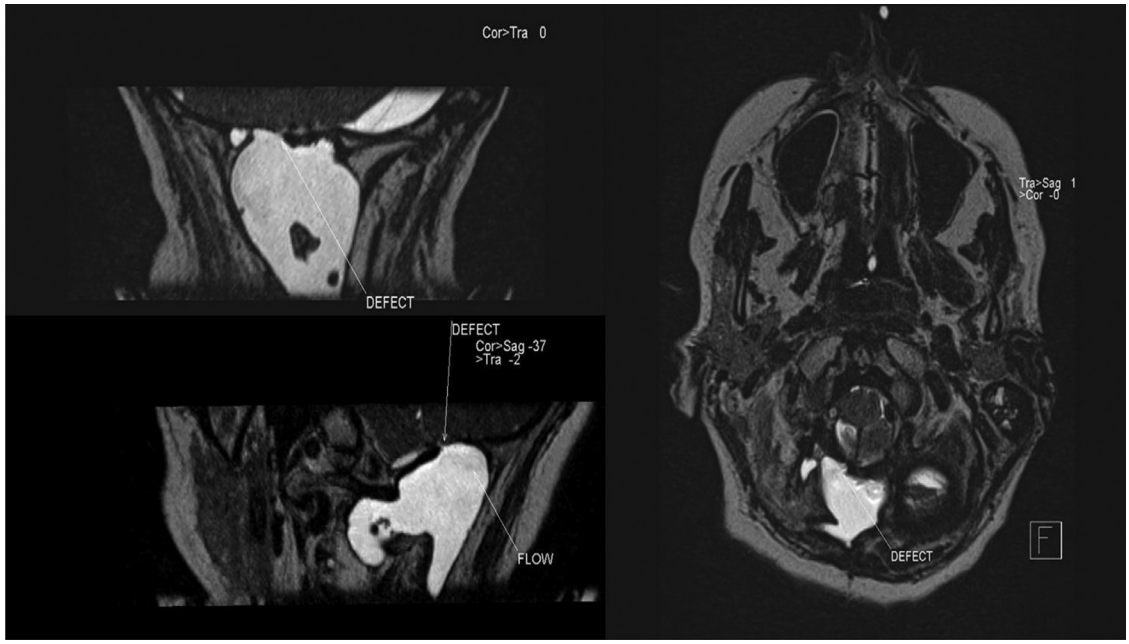

Fig. 2 – In T2 space images, there is a small defect in the dura mater of foramen magnum with CSF jet into extradural space and CSF collection in the subcutaneous region adjacent to the surgical bed.

Fig. 3 – In sagittal T2 sequence, complete disappearance of pseudomengocell in surgical bed and regression of brain sagging.

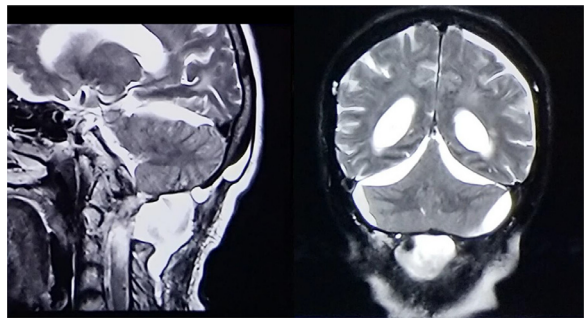

Fig. 4 – For these images in sagittal and coronal view in T2 sequence, defect in dura matter of foramen magnum with CSF jet into extradural space and CSF collection in surgical bed with brain sagging, and cerebellar tonsil ectopia with the expansion of subarachnoid spaces above cerebellar hemispheres are seen.

CSF leakage location. However, it is invasive and may be associated with higher rates of related complications.

Moreover, radioisotope cisternography is no longer recommended [2,4]. Hence, there is an excellent preference for using MR CSF flow studies. In the current case, imaging findings helped diagnose intracranial hypotension, and the author recruited the CSF flow study to determine the exact location of the CSF leak.

Former studies recommended treatment options such as epidural blood patching and fibrin glue injection in the leakage site in the case of spontaneous intracranial hypotension [5–7]. We believe that primary surgical closure of the leakage origin is more effective in the postoperative CSF leak. In the current case, the patient underwent surgical duraplasty and primary closure of the CSF leak site. There was significant clinical and imaging improvement in her 2 years follow-up, and no evidence of recurrence was noted.

Conclusion

Diagnosing IH following the CSF leak during the postoperative phase could be challenging. Due to unspecific signs and symptoms and normal clinical findings, particularly in the initial stages, the diagnosis may be delayed or missed. So, a high

rate of clinical suspicion plays an essential role in diagnosing this condition.

Patient consent

Written informed consent for publication of clinical findings of current case report was obtained from the patints

Authors' contributions

All co-authors read and approved the final manuscript. MS and KE: provided technical insights; MME and HHT: essential manuscript drafting and technical proofreading; MH: contributed to the manuscript editing; SAM and ORM: provided technical insights.

REFERENCES

[1] Forghani R, Farb R. Diagnosis and temporal evolution of signs of intracranial hypotension on MRI of the brain. Neuroradiology 2008;50(12):1025.

[2] Sainani N, Lawande M, Pungavkar S, Desai M, Patkar D, Mohanty P. Spontaneous intracranial hypotension: a study of six cases with MR findings and literature review. Australasian Radiol 2006;50(5):419–23.

[3] Sharifi G, Mousavinejad SA, Bahrami-Motlagh H, Eftekharian A, Samadian M, Ebrahimzadeh K, et al. Delay posttraumatic paradoxical cerebrospinal fluid leak with recurrent meningitis. Asian J Neurosurg 2019;14(3):964–6.

[4] Ferrante E, Prone V, Rubino F, Ferrante M. Spontaneous intracranial hypotension with chronic brain sagging causing foramen magnum CSF circulation disorder reversible after lumbar epidural blood patch. Neurol Sci 2018;39(6):1137–8.

[5] Ferrante E, Arpino I, Citterio A, Wetzl R, Savino A. Epidural blood patch in Trendelenburg position pre-medicated with acetazolamide to treat spontaneous intracranial hypotension. Eur J Neurol 2010;17(5):715–19.

[6] Wu J-W, Hseu S-S, Fuh J-L, Lirng J-F, Wang Y-F, Chen W-T, et al. Factors predicting response to the first epidural blood patch in spontaneous intracranial hypotension. Brain 2017;140(2):344–52.

[7] Franzini A, Messina G, Nazzi V, Mea E, Leone M, Chiapparini L, et al. Spontaneous intracranial hypotension syndrome: a novel speculative physiopathological hypothesis and a novel patch method in a series of 28 consecutive patients. J Neurosurg 2010;112(2):300–6.

[8] Mea E, Chiapparini L, Savoiardo M, Franzini A, Bussone G, Leone M. Clinical features and outcomes in spontaneous intracranial hypotension: a survey of 90 consecutive patients. Neurol Sci 2009;30(1):11–13.

[9] Kranz PG, Gray L, Amrhein TJ. Spontaneous intracranial hypotension: 10 myths and misperceptions. Headache 2018;58(7):948–59.

[10] Kranz P, Tanpitukpongse T, Choudhury K, Amrhein T, Gray L. Imaging signs in spontaneous intracranial hypotension: prevalence and relationship to CSF pressure. Am J Neuroradiol 2016;37(7):1374–8.

[11] Kranz PG, Tanpitukpongse TP, Choudhury KR, Amrhein TJ, Gray L. How common is normal cerebrospinal fluid pressure in spontaneous intracranial hypotension? Cephalalgia 2016;36(13):1209–17.

[12] Pulliam Z, Somers A, Strazis C. Iatrogenic intracranial hypotension: a case study. J Radiol Nurs 2020;39(4):327–30.