



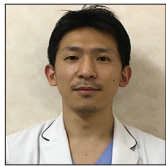
Case Report

The algorithm for diagnosis and management of intracranial hypotension with coma: Report of two cases

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Received : 22 July 2020

Accepted : 07 August 2020

Published : 29 August 2020

DOI

10.25259/SNI_460_2020

Quick Response Code:



ABSTRACT

Background: Spontaneous intracranial hypotension (SIH) is caused by spontaneous cerebrospinal fluid (CSF) leaks that can be treated in most cases with an epidural blood patch (EBP). However, some patients, who develop severe brain sagging, can neurologically deteriorate, and in occasional instances, which become comatose. Here, with the presentation of two cases, and a review of the literature, we have set guidelines for diagnosing SIH along with recommendations for its management.

Case Description: We reviewed two cases of SIH. Both patients became comatose due to a CSF leak associated with a tear in the spinal dura diagnosed on myelo-CT studies. As targeted EBP failed to achieve sustained improvement, direct operative repair of the dural tears was warranted (video presentation).

Conclusion: After reviewing two cases of SIH and the literature, we developed an algorithm for the diagnosis and management of SIH. To avoid deterioration to a comatose status, we recommend the early performance of myelo-CT studies to identify the location of the dural leak, followed by early dural repair.

Keywords: Cerebrospinal fluid leak, Coma, Dural repair, Epidural blood patch

INTRODUCTION

Spontaneous intracranial hypotension (SIH) commonly causes orthostatic headaches. The standard treatment is initial conservative management utilizing an epidural blood patch (EBP).^[1-3] However, in rare cases, patients can neurologically deteriorate to the point of becoming comatose due to “severe brain sagging.”^[1-3] Here, we reviewed two new cases of SIH where patients presented in a coma and followed with a targeted review of the literature.

CASE REPORTS

Case 1

A 52-year-old woman presented with persistent orthostatic headaches and was diagnosed with SIH. She underwent an MRI that demonstrated deformation/severe sagging of the midbrain with bilateral subdural hematomas [Figure 1a and b]. As a spinal CSF leak was suspected, she was scheduled to receive an EBP. However, on the second hospital day, her GCS score acutely deteriorated to 8 (E2V1M5), and she developed bilateral miosis (right, 2 mm and left, 2 mm) with

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bradycardia. After being placed in the Trendelenburg position (-10°), her GCS score improved back to 15 within 1 day.

Myelo-CT study

The myelo-CT study revealed a very low opening pressure of just 4 cm H₂O, while the myelo-CT itself revealed a micro ventral spur and dural tear at the Th11/12 level [Figure 1c and d]. A targeted T11/T12 EBP with bilateral hematoma drainage resulted in immediate improvement back to a GCS score of 15. She was discharged home 13 days after the original admission. However, 9 days postdischarge, the left subdural hematoma, and brain sagging recurred. Following a Th11/12 laminotomy, a large dural tear (8 × 2 mm) on the ventral aspect of the dura mater was repaired with a fat graft (video presentation). In retrospect, muscle grafting is a preferred technique as it retains its size/configuration, whereas fat grafts typically resorb/shrink resulting in more recurrent CSF leaks. One year postoperatively, she continued to do well.

Case 2

A 61-year-old man with orthostatic headaches was transferred due to altered mental status. The brain MRI

also showed deformation of the mesencephalon-midbrain due to severe brain sagging along with bilateral thick subdural hematomas. As a spinal CSF leak was suspected on the cervicothoracic MRI [Figure 1e], a myelo-CT was performed. When the myelo-CT demonstrated a microspur with dural leak ventrally at the T1/2 level; a targeted EBP was performed. Eighteen days later, when the orthostatic headaches and brain sagging recurred on the subsequent brain MRI, the patient underwent T1/T2 microspur removal with dural repair [Figure 1f-h and Table 1]. Six months postoperatively, the patient continued to do well.

DISCUSSION

In general, SIH often resolves with conservative treatment, particularly with the performance of EBP.^[5] However, in rare cases, patients with SIH can demonstrate rapid neurological deterioration resulting in coma attributed to cerebral herniation.^[1-3] Here, we presented two cases where SIH was attributed to spinal dural tears that failed to resolved with targeted EBP. In both cases, patients required direct spur removal/dural repairs of their CSF fistulas.

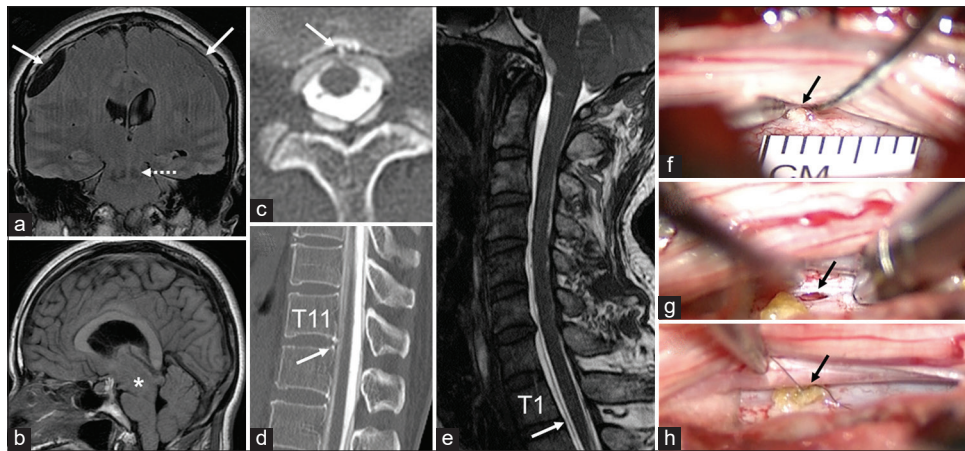


Figure 1: Case 1: Brain MRI on admission showed bilateral subdural hematoma (a: arrows), downward shifting of the red nucleus (a: dotted arrow), and deformation of the diencephalon-midbrain due to severe brain sagging (b: asterisk). Myelographic CT showed microspurs on the ventral side of the dura at the Th11/12 level (c: arrow) and a leak of the contrast medium out of the dura (d: arrowhead). Case 2: A spinal CSF leak was suspected in spine MRI (e: arrow). A dural tear (2 × 1 mm) and a microspur were found in the ventral dura at the Th1/2 level (f: arrow). The microspur was removed (g: arrow) and the dural tear was sealed with subcutaneous fat tissue (h).

Table 1: Summary of our cases of spontaneous intracranial hypotension with coma.					
Case number	Age/sex	Lowest GCS score	Leak site	Treatments	Outcome
1	52/F	8	Th11-12	1. Targeted EBP (T11/12) 2. Dual repair	GR
2	62/M	6	Th1-2	1. Targeted EBP (T2/3) 2. Dural repair	GR

GCS: Glasgow Coma Scale, GR: Good recovery, EBP: Epidural blood patch

Treatment alternatives for SIH

There are a variety of methods for treating SIH. Nonsurgical treatment, includes nontargeted lumbar EBP,^[3] targeted EBP,^[4] epidural fibrin glue patch, and/or intrathecal saline infusion.^[1] The main surgical alternative is the direct dural repair of spinal spurs/CSF fistulas; this was performed in this case and reported in four prior SIH cases involving comatose patients.^[1-4]

Algorithm for the diagnosis and management of SIH

Here, we propose an algorithm for the diagnosis and management of SIH [Figure 2]. When a coma is recognized, the patient should be placed in the Trendelenburg position; this should be followed by a targeted EBP. However, after multiple EBP have been performed and are unsuccessful, early adoption direct spur removal/dural repair is warranted. The latter direct techniques should avoid long-term permanent neurological sequelae observed when, alternatively, patients have continued to undergo nonsurgical management/ “observation” alone.

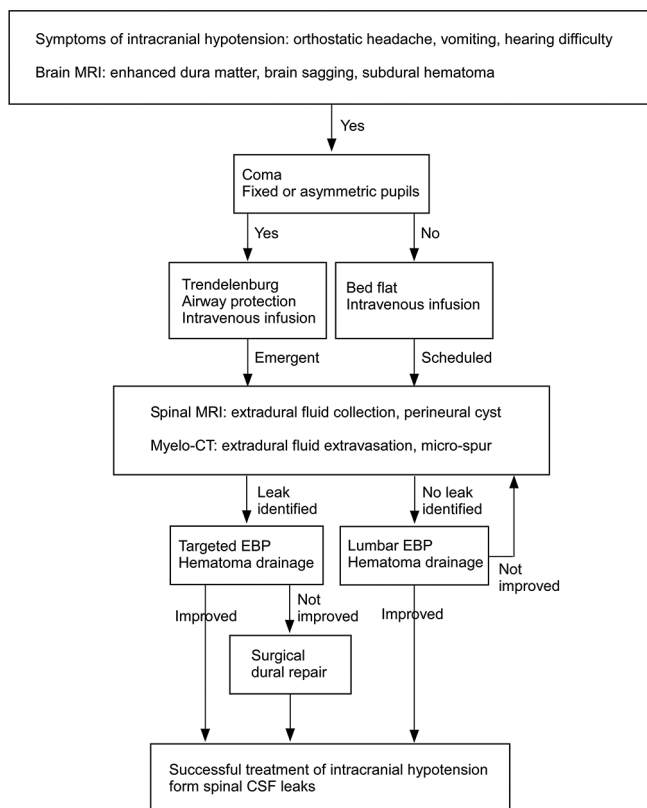


Figure 2: The algorithm for diagnosis and management of intracranial hypotension with coma.

CONCLUSION

The clinical decision-making algorithm presented here may help neurologists/neurosurgeons who diagnose and treat this rare condition.

Acknowledgments

Dr. Shintaro Arai and Dr. Keisuke Takai contributed equally to this work and share first authorship.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Arai S, Takai K, Taniguchi M. The algorithm for diagnosis and management of intracranial hypotension with coma: Report of two case. *Surg Neurol Int* 2020;11:267.