Interdisciplinary Neurosurgery xxx (xxxx) xxx–xxx

Contents lists available at ScienceDirect

Interdisciplinary Neurosurgery

journal homepage: www.elsevier.com/locate/inat

Case Report

Spontaneous intracranial hypotension resulting in coma: Case report and review of the literature☆

Zamir Merali, MD[a], Christopher D. Witiw, MD, MS[a], Shelly Wang, MD, MPH[a], Nicholas Phan, MD, FRCSC[b], Victor Yang, MD, PhD, PEng FRCSC[b,*]

[a] Division of Neurosurgery, Department of Surgery, University of Toronto, Toronto, Ontario, Canada
[b] Division of Neurosurgery, Department of Surgery, Sunnybrook Health Sciences Center, Toronto, Ontario, Canada

ARTICLE INFO

Keywords:
Spontaneous intracranial hypotension
Neurosurgery
Subdural hematoma

ABSTRACT

Spontaneous intracranial hypotension (SIH) has a wide range of clinical presentations. Cases of SIH that result in severe neurologic compromise have only very rarely been described.

We describe our experience with a 70-year-old male who presented with a history of postural headache and imaging that demonstrated bilateral subdural hematomas. The patient's condition subsequently deteriorated and he became comatose despite an epidural blood patch and craniotomy to drain the subdural hematoma. We utilized a strategy of intraventricular saline infusion to normalize intracranial pressure. The patient ultimately required direct surgical ligation of a spinal perineural cyst before making a full recovery.

Given the rarity of SIH presenting with severe neurologic deterioration and the unique treatment strategies required to manage this condition we conducted a review of the literature and discussed the management of the most severe complications of SIH.

1. Introduction

Spontaneous intracranial hypotension (SIH) is a differential consideration in patients presenting with orthostatic headaches, non-traumatic spontaneous subdural hematomas, and neurologic decline. Since the original description of SIH in 1938, an understanding of the pathophysiology of the disease has been improved by contrast-enhanced magnetic resonance imaging (MRI) and treatment strategies have become established [1]. Conversely, cases of SIH that result in severe neurologic compromise have only very rarely been described, and treatment strategies are not well established. Here we present a case of SIH complicated by severe neurologic deterioration where a treatment strategy of intraventricular saline infusion was effectively applied. We discuss the diagnosis and treatment of SIH and present treatment strategies for the management of the most severe complications of this condition.

2. Case report

A 70-year-old Caucasian male with a remote history of unprovoked pulmonary embolus for which he took warfarin presented with a 9 day history of postural headache that was accompanied by increasing confusion. At admission he had a Glasgow Comma Scale (GCS) score of 14, with a 1-point decrement for disorientation and no focal neurologic deficits. A brain computed tomographic (CT) scan demonstrated bilateral subdural hematomas with the left-side measuring approximately 1.2 cm and the right-side 0.9 cm and 1 cm of mid-line shift.

Given the history of postural headaches that were worse when upright and improved with recumbancy and the finding of bilateral subdural hematomas, the diagnosis of intracranial hypotension was considered. The patient was placed on strict bed rest and his warfarin was held. A magnetic resonance imaging (MRI) scan of the brain demonstrated again large bilateral subdural hematomas (Fig. 1A). In addition, generalized patchy-meningeal thickening and enhancement was noted along with minimal effacement of the basal cisterns and downward displacement of the midbrain and brainstem onto the clivus (Fig. 1B). An MRI of the entire spine was completed and showed a thin posterior epidural collection extending from T11-L5. In addition, a perineural cyst was identified at T11/12 on the left side (Fig. 1C,D).

Due to the patient's progressive neurologic decline and MRI findings

Abbreviations: SIH, Spontaneous intracranial hypotension; MRI, Magnetic Resonance Imaging; GCS, Glasgow Comma Scale; CT, Computed Tomography; EVD, External Ventricular Drain; ICP, Intracranial Pressure

☆ **Disclosures**: the authors have no conflicts of interest to disclose, and no financial relationships relevant to this article to disclose.

* Corresponding author at: Department of Surgery, Sunnybrook Health Sciences Center, M6 100|2075 Bayview Ave., Toronto, Ontario M4N 3M5, Canada.
E-mail address: victor.yang@sunnybrook.ca (V. Yang).

http://dx.doi.org/10.1016/j.inat.2017.08.009
Received 19 July 2017; Received in revised form 9 August 2017; Accepted 27 August 2017

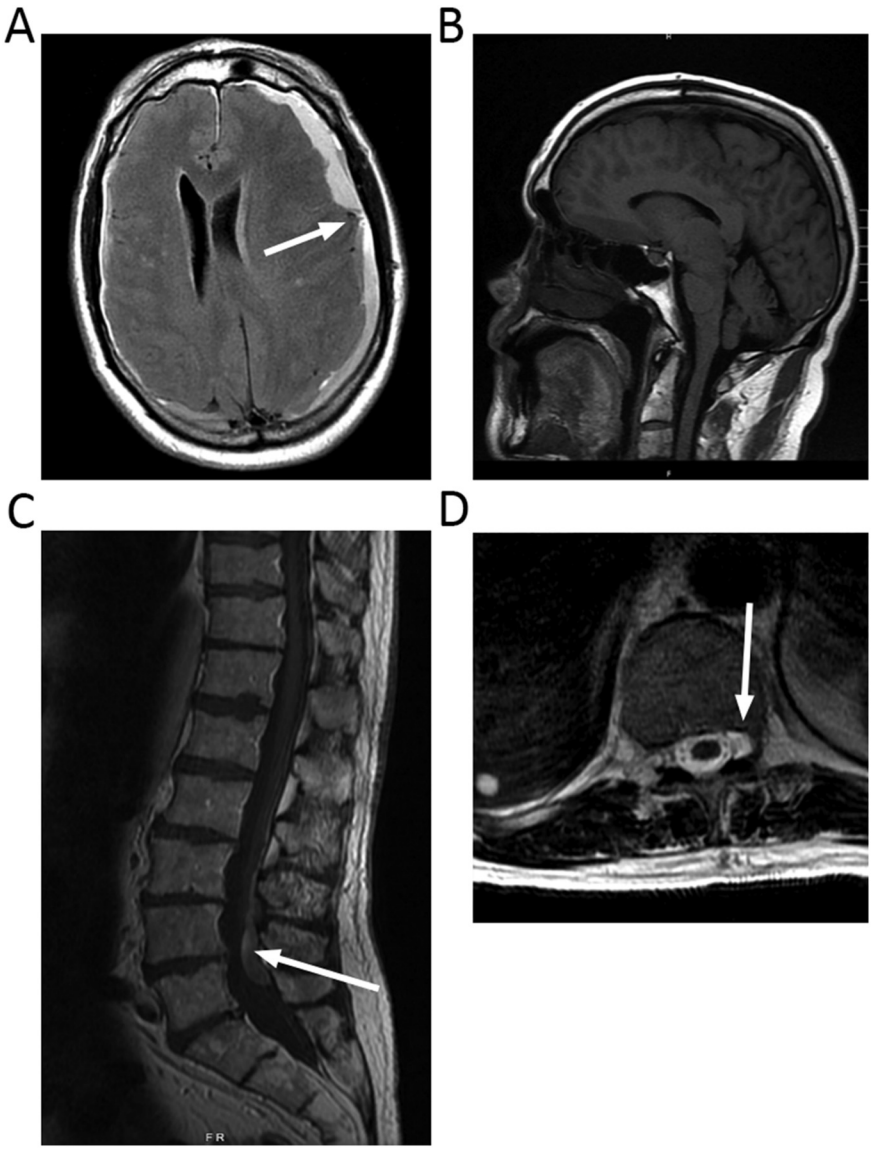

Fig. 1. Initial imaging investigations. T1 weighted contrast-enhanced MRI (A) demonstrating bilateral subdural hematomas, midline shift, and descent of the midbrain onto the clivus (B) consistent with spontaneous intracranial hypotension. A posterior epidural collection was seen in the lower lumbar spine (C) and a perineural cyst was present in the left neuronal foramen at T11/12 (D).

suggestive of intracranial hypotension with a potential spinal CSF leak, we consulted the anesthesia service for emergency injection of a lumbar epidural blood patch.

On the 9th day post-admission the patient deteriorated to a GCS score of 11. He became non-verbal and localized only to painful stimuli. An emergency CT scan demonstrated that the left-sided subdural hematoma had expanded to 1.9 cm with 1.7 cm of mid-line shift and left-sided uncal herniation. With this sudden neurologic decline and concerning radiographic findings we emergently brought the patient to the operating room for a left-sided craniotomy and evacuation of the subdural hematoma. A mutli-septated subdural hematoma with chronic and acute components was evacuated and a watertight duraplasty was completed with synthetic dural matrix.

Following surgery the patient was extubated and transferred to the intensive care unit. An immediate post-operative CT scan demonstrated that the left subdural collection had been evacuated and the mid-line shift and uncal herniation had partially resolved. Despite this the patient remained in poor neurologic condition and we therefore performed another CT scan that demonstrated the left subdural hematoma had re-accumulated to a thickness of 1.7 cm.

At this time the patient underwent a thoracic epidural injection of 10 mL of autologous blood and was then kept in Trendelenburg position of 10 degrees. After the blood patch the patient's neurologic status initially improved and he was able to open his eyes spontaneously and localize to pain. Unfortunately, after this transient improvement, the patient's clinical status subsequently worsened and he required intubation for airway protection due to his declining neurological status.

At this time, we placed an external ventricular drain (EVD) in the right lateral ventricle for intra-cranial pressure (ICP) monitoring. As expected ICPs were persistently low, ranging between −1 and 4 cmH2O. We also inserted a lumbar epidural catheter and began infusing Tetrastarch into the epidural space.

On the 20th day post-admission the patient's neurologic exam deteriorated suddenly and his only response to painful stimuli was a weak extensor response. At this time the ICP rose suddenly from 2 to 27 cmH2O and the left pupil became fixed and dilated. A CT head confirmed our suspicion that the left subdural hematoma had worsened with evidence of acute blood (Fig. 2A). An emergent left craniotomy was performed to drain the subdural hematoma and an intra-parenchymal ICP monitor was inserted.

Immediately following the operation the ICP was 0–2 cmH2O. With the aim of increasing the ICP and localizing the source of the CSF leak we infused an 18 mL mixture of 1:1 Iohexol and saline through the right frontal EVD over the course of 2 h. During this infusion the ICP rose to 8-12cmH_2O and remained stable at this level after the infusion was stopped and the EVD was clamped (Fig. 3). An immediate CT head