Clinical Neurology and Neurosurgery 111 (2009) 699–702

Contents lists available at ScienceDirect

Clinical Neurology and Neurosurgery

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

Case report

Coma resulting from spontaneous intracranial hypotension treated with the epidural blood patch in the Trendelenburg position pre-medicated with acetazolamide

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ARTICLE INFO

Article history:
Received 30 July 2008
Received in revised form 31 May 2009
Accepted 2 June 2009
Available online 3 July 2009

Keywords:
Headache
Spontaneous intracranial hypotension
Subdural haematoma
Coma
Epidural blood patch
Acetazolamide

ABSTRACT

A 62-year-old man had a new onset of severe, orthostatic headache which eventually progressed to a stupor and a coma 3 weeks later. A computed tomography (CT) scan showed bilateral chronic subdural haematoma and magnetic resonance imaging (MRI) of the brain showed the typical findings of spontaneous intracranial hypotension (SIH). After pre-medication with acetazolamide, he was treated with three lumbar autologous epidural blood patches (EBPs) and kept in the Trendelenburg position, with full recovery. The first lumbar autologous EBP was ineffective and the second was only partially effective because of incorrect execution of the procedure as shown by spinal neuroimaging examination post-EBP. A spinal neuroimaging examination post-EBP is therefore to be recommended in order to confirm the correct execution of procedure. Pre-medication with acetazolamide and keeping the patient in the Trendelenburg position could reduce the flow of spinal cerebrospinal (CSF) leak favouring sealing of the hole.

1. Introduction

Spontaneous intracranial hypotension (SIH) is characterized by orthostatic headaches, low CSF pressure and distinct abnormalities on MRI [1,2]. It generally results from spinal spontaneous CSF leakage, sometimes associated with underlying connective tissue disorders [3]. Treatment ranges from conservative management, such as bed rest, over-hydration and caffeine, to invasive procedures, such as the autologous epidural blood patch (EBP) [4–6], CT-guided fibrin glue injection at the site of the leak [7], and open surgical intervention. EBP has emerged as the treatment of choice for SIH when initial conservative measures fail to bring relief. Sencakova et al. [4] reported 25 cases of SIH treated with EBP, and found that 36% of all patients recovered with one EBP. Berroir et al. [5] observed a 57% success rate in a series of 30 patients with SIH after one EBP. We described 32 patients with SIH and observed a 90% success rate after one EBP [8]. SIH may rarely cause coma, and in these cases the choice of treatment is very challenging [9]. We report a patient with coma from SIH who recovered following repeated lumbar autologous EBPs pre-medicated with acetazolamide.

2. Case report

A 62-year-old man, treated with warfarin for a mechanic aortic valve prosthesis, developed severe, diffuse orthostatic headache with nausea and vomiting. There was no history of connective tissue disorders, alcoholism or trauma. After 15 days he became progressively obtunded. Laboratory test results showed INR of 2.3 (normal 0.8–1.3). This value was within therapeutic levels, and there were no metabolic disorders. Haematological and biochemical laboratory tests, monitoring of thyroid hormones and inflammation rate were normal. He was admitted to the neurosurgical department. Brain CT revealed small bilateral chronic subdural haematomas (SH), increased attenuation in the sylvian fissures (Fig. 1). Cerebral angiography was normal. He underwent an evacuation of bilateral chronic SH because they were erroneously considered by neurosurgeons to be the cause of obtunding. Two days later the patient became comatose, the Glasgow coma scale score (GCSS) was 5 (eye opening response 1, motor response 3, verbal response 1). He showed respiratory distress and was intubated for 10 h. The patient again developed two episodes of respiratory distress with intubation the third and sixth days after the operation. Afterwards the neurology specialist suspected SIH, advised cerebral RMN with gadolinium.

Brain MRI, 12 days after operation, showed diffuse pachymeningeal enhancement and brain sagging (Fig. 2A). These features

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doi:10.1016/j.clineuro.2009.06.001

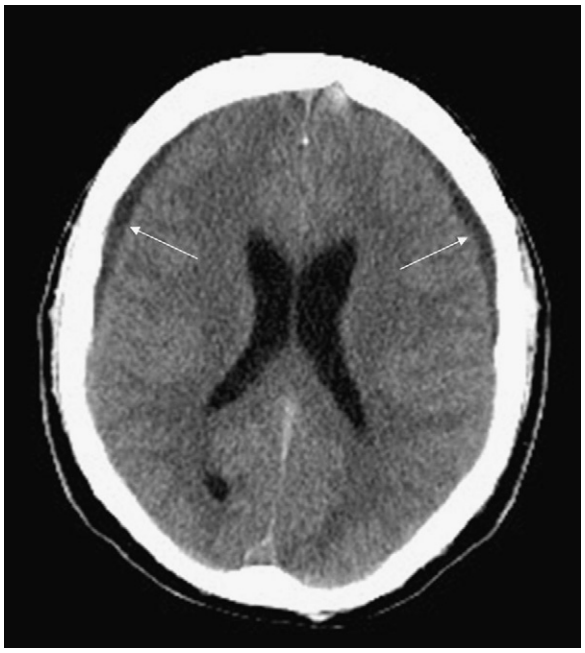

Fig. 1. Axial CT scan without contrast shows small chronic subdural haematomas interesting both frontal regions (white arrows).

were considered to be consistent with SIH and severe resultant diencephalic compression. Spinal and myelo-MRI failed to prove a CSF leak. He was placed in the Trendelenburg position at about 30° and awoke within 8 h, GCSS 15 [E4 M6 V5]. The next day the patient underwent a lumbar autologous EBP with 35 ml of blood mixed with 1 ml of gadolinium. The EBP was performed at L4–L5 level with a 16-g Tuohy needle via a midline approach with the patient lying on his left side. The epidural space was identified with the loss of resistance technique using air. The injection was stopped when the patient reported an increased pressure sensation in the neck. Spinal MRI post-lumbar autologous EBP showed that only a small quantity of blood went into epidural space at L5–S1 level whereas considerable blood was localized into paraspinal musculature (Fig. 3A). Twenty-four hours after lumbar autologous EBP, when the patient was in anti-Trendelenburg position at about 45°, he again became stuporous and inattentive. Another lumbar autologous EBP at L4–L5 level was therefore given under fluoroscopy guidance with 30 ml of blood mixed with 5 ml of iopamidol with patient lying in prone position. A multislice spiral spinal CT post-lumbar autologous EBP showed small quantity of air in the epidural space from L5 to D11 (Fig. 3B) and blood mixed with contrast medium was localized only in paraspinal musculature. The patient progressively improved over the next 15 days. He then again became progressively stuporous, asthenic, and was affected by disphagia. We performed another lumbar autologous EBP at L2–L3 level under fluoroscopy guidance (27 days after the second EBP) with 30 ml of blood mixed with 5 ml of iopamidol. Spinal CT post-EBP showed blood in epidural space from L1 to C7–D1 level (Fig. 3C). After 24 h the headache disappeared and he improved. He had rapid recovery of function soon after the third EBP.

Oral anti-coagulant therapy with warfarin was stopped until INR was normal and before EBP treatment. Meantime enoxeparine 0.4 ml was administrated subcutaneously, twice per day. Warfarin treatment was resumed after EBPs. The patient was treated with a pre-medication with acetazolamide at dosage of 500 mg 12 h before giving lumbar autologous EBPs. He was kept in a 30° Trendelenburg position since an hour before the procedure, during the procedure and 24 h after the EBPs. Bilateral chronic SH were resolved completely after the first surgery. Brain MRI was normal after 4 months (Fig. 2B). After 24 months of follow-up the patient was in good health, he had no clinical sequelae and limitations related to the episode. He was fully functional, he was on warfarin and he had no recurrences.

3. Discussion

In this case bilateral chronic SH were caused by SIH by tearing of bridging veins resulting from the downward displacement of the brain, the coma by severe sagging of the brain leading to diencephalic deformation and dysphagia by bulbar weakness to deep midline structure [10]. SH could been attributed solely to the warfarin use and the obtunding, incorrectly, to the SH, but the INR on therapeutic range and the positional feature of the headache suggested the diagnosis of SIH. The SH in SIH are frequently small and may be asymptomatic, but sometimes become large and symptomatic, cause notable mass effect and shift the midline structures. Readers may wonder whether the fact that the patient was on warfarin may have contributed to the bleed and although this must always be a consideration, we do not think this was a contributing factor in this case. In fact, we had previously observed six patients with SIH complicated with bilateral chronic SH with mass effect

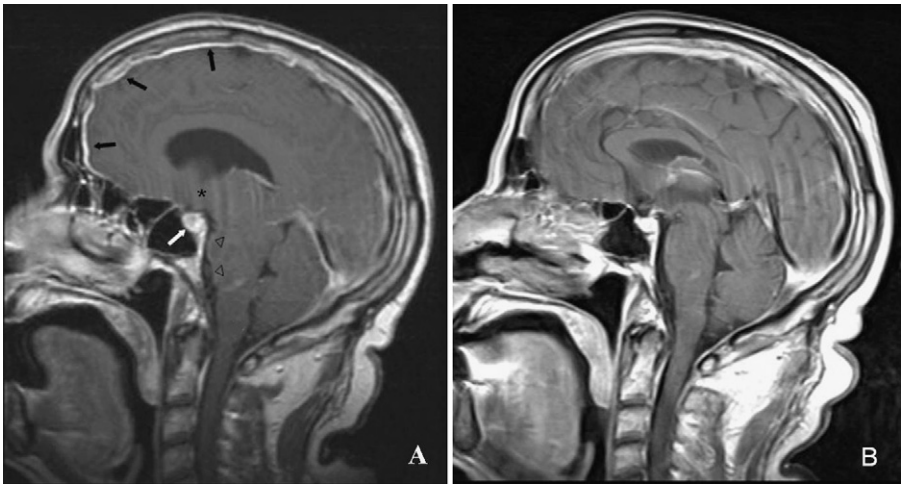

Fig. 2. Sagittal T1-weighted brain MRI with gadolinium showing diffuse pachymeningeal enhancement (black arrows), pituitary gland enlargement (white arrow), flattening of the pons with partial obliteration of the basilar cisterns (black open arrowheads), descent of the hypothalamic structures (asterisk) (A); sagittal T1-weighted brain MRI with contrast acquired 4 months after the EBP procedure showing resolution of the previously described signs of SIH (B).