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Case report

Postoperative intracranial hypotension-associated venous congestion: Case report and literature review

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1. Introduction

Postoperative intracranial hypotension-associated venous congestion (PIHV), formally proposed in 2003 by Van Roost et al. as pseudohypoxic brain swelling, is a rapid, severe, and potentially fatal postoperative complication following uneventful intracranial or spinal surgery [1,2]. PIHV is characterized by unexpected postoperative deterioration of consciousness; neuroradiological changes in the basal ganglia, thalamus, brainstem, and cerebellum resembling the radiological presentation of hypoxic brain damage; and cerebral arteries and veins that remain angiographically intact with possible neuroimaging signs suggesting intracranial hypotension [2,4]. PIHV is thought to be caused, pathomechanistically, by intracranial hypotension as a result of subfascial or subgaleal drainage in patients whose dural sutures allow for cerebrospinal fluid (CSF) permeation [1–3].

The mortality of PIHV has yet to be fully described in literature, and as a whole this complication remains subjective among surgeons despite the large number of well documented cases [1–3,5–7].

We present a case of death following the development of PIHV from the lowest amount of CSF loss yet reported, during an otherwise uneventful bone flap replacement using a custom made cranioplastic bone graft, and analyze PIHV which results in death, while highlighting a potential strategy for avoidance.

2. Case report

A 46-year-old male, with no significant past medical history, presented in December of 2008 with thrombosis of the right middle cerebral artery and underwent a decompressive craniectomy. In January of 2009 the patient underwent surgery to place an autologous bone graft and a ventriculoperitoneal (VP) shunt for the management of hydrocephalus. Good cognitive status, left hemiparesis, and slight anisocoria were observed during post-operative recovery in the intensive care unit.

In February of the same year, the patient fell while undergoing rehabilitation resulting in an injury to the bone flap. The bone flap shifted from its placed position and sank into the original surgical opening. This was fostered, in part, by resorption of the bone flap. Computed tomography (CT) showed no new lesions or any signs of CSF overdrainage (Fig. 1).

The patient requested that a custom made 3D CT-based cranioplastic bone graft be placed. This was performed in December of 2010. Surgical placement was uneventful and anesthesia was maintained with sevoflurane. After making a skin incision along the original scar, the diastatic bone flap was removed and the dura

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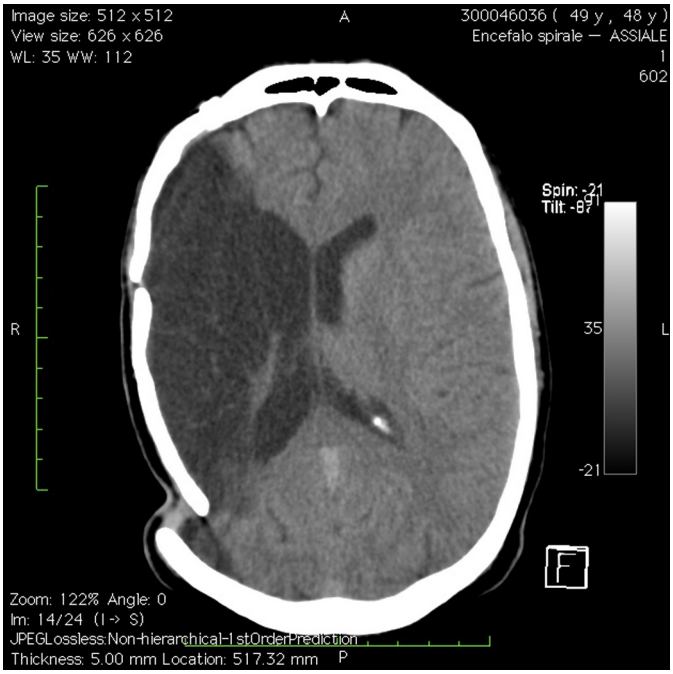

Fig. 1. Preoperative axial CT showing detachment and sinking of a right autologous bone flap. Hypodense regions of the right hemisphere from previous middle cerebral artery thrombosis are visible.

was gently detached from the bone. No swelling was observed during the procedure. The existing VP shunt was inspected and left in situ. The valve-regulator was confirmed to have a pressure setting of 120 mmH_2O. Accidentally, a small dural perforation was made and the outflow of cerebrospinal fluid was approximately 15 cm^3. The new polymethylmethacrylate cranioplastic prosthesis was implanted into the opening, and it fit well. The surgery ended with dural suspension and skin suturing. A subgaleal drain was placed, low negative pressure suction (approximately −150 mmHg) was applied, as was routine, and within several seconds approximately 50 cm^3 of CSF was drained. Upon observing this amount of CSF drainage, vacuum suction was instantly discontinued.

Immediately postoperative the patient developed pupillary fixation and dilation, and severe diffuse bilateral cerebral edema was observed on CT (Fig. 2). CT angiography showed an intact Willis polygon with no arterial occlusions (Fig. 3). Blood chemistries, including electrolytes and a coagulation profile were unremarkable. The endotracheal tube was correctly positioned, as confirmed by CT, and recorded oxygen saturation values remained above 98% at all times. The patient was cardiovascularly stable throughout the procedure, with mean arterial pressure values between 75 and 90 mmHg recorded. The patient was pronounced dead approximately 48 h post-surgery.

3. Discussion

Pseudohypoxic brain swelling was first defined in 2003, as a surgical complication whose clinical features mimic global cerebral hypoxia, by Van Roost et al. as part of a comprehensive investigation on a series of 17 patients [1]. Through their investigation they were able to rule out anoxemic and ischemic hypoxia, respiratory chain inhibition, mitochondriopathy, poisoning, and adverse drug reactions as causes; and concluded that this complication is the result of intracranial hypotension caused by the application of subgaleal suction drainage [1].

In 2010, Hadizadeh et al. reviewed radiological findings associated with the previous series along with additional cases and found signal intensity changes in the basal ganglia, prompting them to redefine this complication as postsurgical intracranial hypotension [3].

Parpaley et al., in 2011, reported the first two cases of this complication following spinal surgery and proposed a more in-depth pathomechanism [2]. They proposed that this severe diffuse cerebral swelling is due to sudden exacerbation of intracranial hypotension caused by subgaleal or subfascial suction drainage which, in accordance with the Monro–Kellie doctrine, induces venous congestion subsequently leading to venous infarction [2,3,5,8]. They thus re-proposed this complication as postoperative intracranial hypotension-associated venous congestion [2].

We reviewed 21 cases reported in the previous series, collected from multiple institutions, and assessed them along with the case we report here (Table 1) [1–3,9]. We analyzed 6 of those cases, including the one herein, which resulted in death following rapid CSF loss through subfascial or subgaleal suction drainage after uneventful intracranial or spinal surgery with dural opening. 6 patients, 4 male and 2 female, with a mean age of 46 years (range, 32–71 years) were reviewed.

In these cases, the mean amount of subfasical/subgaleal drainage volume was 365 ml (standard deviation, 159 ml; range, 50–710 ml; 8 cases reported no data). The Monro–Kellie doctrine provides credence to the theory that PIHV is the result of CSF hypovolemia, and states that CSF loss would require intracranial volume compensation by blood or brain [8]. Thus, compensation should occur through an increase in cerebral blood volume, especially venous blood volume as veins are much more distensible than arteries [3,6,10]. This increase in venous blood volume could induce venous congestion, and in some cases subsequently venous infarction, concomitant with vasogenic and cytotoxic edema [3,10]. Brain sag as a result of CSF hypovolemia, and the resulting compression of the deep venous system, are additional pathomechanistic possibilities, although to date only angiographically negative findings have been reported – aside from slow circulation time which could non-specifically indicate microvascular dysfunction secondary to cerebral edema [1,9]. Though it has been ruled out in a number of cases, magnetic resonance findings in one reported case were similar to those of deep cerebral venous thrombosis, and as spontaneous intracranial hypotension has been shown to be a risk factor for cerebral venous thrombosis, this pathomechanistic possibility cannot yet be completely ruled out [7,9].

In a considerable subset of the reviewed cases, 29% (6 of 21 patients), PIHV resulted in death. This subset of cases had a mean subfasical/subgaleal drainage volume of 268 ml (range, 50–390 ml), and mean time after surgery until death of 48 h (range, 25–72 h). The cases of non-fatal PIHV had a mean subfasical/subgaleal drainage volume of 402 ml (standard deviation, 150 ml; range, 170–710 ml; 8 cases did not report this data), suggesting that fatal PIHV is not a factor of the quantity of CSF loss but possibly of the velocity of CSF outflow – itself a factor of the level of the negative pressure applied. Past surgical history may also play a role. In all 21 cases of PIHV, 33% (7 of 21 patients) had previous cranial surgery, and of the patients who developed fatal PIHV, 67% (4 of 6 patients) had previous cranial surgery.

Due to this high mortality rate, we use the term postoperative intracranial hypotension-associated death (PIHD) to describe cases of fatal PIHV.

The case of PIHD described here is the lowest reported loss of CSF volume (50 cm^3) that has resulted in the development of PIHV, and the second known case of PIHD, and PIHV, in a patient with an existing VP shunt. This is also the only known case where sevoflurane was used as the anesthetic agent, though this is likely an incidental and non-contributory finding. Previously, the lowest