Oculomotor Palsy in Spontaneous Intracranial Hypotension: Case Report and Review of the Literature

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Pathological loss of cerebrospinal fluid (CSF) through small leaks in the spinal meninges causes the typical postural headache associated with spontaneous intracranial hypotension (SIH). The most common causes are iatrogenic, notably secondary to lumbar puncture or spinal surgery. Commonly associated symptoms are neck stiffness, vertigo, nausea and vomiting, and tinnitus, as well as ophthalmoplegia. To our knowledge, eight cases of oculomotor nerve palsy associated with SIH are reported in the literature. Three cases of patients on anticoagulation with subdural haematomas secondary to SIH are reported. Herein we report the first case of painful oculomotor nerve palsy secondary to SIH in a patient on lifelong anticoagulation presenting with subdural haematomas.

We present the case of a 56-year-old Asian woman known for essential thrombocytosis, bilateral mastectomies for breast cancer and deep venous thromboembolism (DVT) with lifelong anticoagulation on warfarin. She was admitted following sudden severe headaches associated with nausea and vomiting as she stood up. Initial computed tomography (CT) scan showed bilateral acute on chronic subdural haematomas (Figure 1). An initial laboratory test showed a prolonged prothrombin time (INR) of 5.2. After standard management of elevated INR in case of bleeding, the follow-up INR was 1. On the third day after admission, she was found to have a partial left third nerve palsy. On further questioning, her headaches had a clear postural component. She had no history of trauma, lumbar puncture or spinal surgery. Examination was normal except for left non-pupil-sparing painful partial third nerve palsy.

A repeat CT head showed stable chronic subdural haematomas and sagging of the brain (Figure 1A,B). CT angiogram revealed no abnormalities. Magnetic resonance imaging (MRI) of the brain (Figure 1C,D) showed such features of intracranial hypotension as diffuse bilateral convexity, dural thickening and enhancement, downward displacement of the midbrain through the incisura with decreased pontine–midbrain angle and distended dural sinuses. Cerebellar tonsils were low-lying. MRI of her entire spine with short tau inversion recovery (STIR) sequences did not identify a clear spinal CSF leak.

The patient was initially put on bed rest in the Trendelenburg position with intravenous fluids. She underwent a successful lumbar epidural autologous blood patch. Her headache and oculomotor palsy improved gradually over the next week. On repeat CT scan after treatment, brain sag was greatly improved (Figure 2).

During hospitalization, the patient developed pleuritic chest pain. She was found to have multiple pulmonary emboli. An inferior vena cava filter was inserted. Upon discharge from hospital, headaches were absent and her painful ophthalmoplegia was close to 95% resolved. Before discharge, a prophylactic dose of dalteparin was started without any increase in size of the subdural hematoma or acute bleeding.

To our knowledge, this is the first case of painful oculomotor nerve palsy secondary to SIH in a patient on anticoagulation presenting with subdural haematomas. CSF plays a suspensor role for the cranial content, preventing downward traction. With loss of CSF in SIH, structures are subject to traction. Most commonly, loss of CSF is due to such iatrogenic causes as lumbar puncture. Traumatic causes also have to be excluded for the intracranial hypotension to be considered spontaneous. Pathophysiology is thought to be due to microruptures of the dura at weak points along spinal root sleeves, through perineural cysts. Traction on cortical or bridging veins can cause subdural haematomas. Downward displacement of the brainstem with compression may cause patients to present in coma. The preferred treatment is autologous epidural blood patches. The level of spinal dural leak does not have to be known for the blood patch to be effective. In comatose patients, lumbar infusion of saline may be needed to improve the level of consciousness while the epidural blood patch is arranged.

Typically, SIH presents with postural headaches. About 12% of patients can present with visual deficits other than diplopia, such as visual acuity and field deficits, nystagmus and photophobia. Associated ophthalmoplegia has been reported in about 30–35% of cases, with the abducens nerve being most frequently implicated in 83% of cases. This may be explained by the sixth cranial nerve’s prolonged intracranial course along the clivus and through Dorello’s canal as well as attachment to the Gruber ligament. A review of the pertinent literature on oculomotor palsy in SIH revealed eight reported cases. One bilateral third nerve palsies were reported in two patients. Two patients presented with both oculomotor and trochlear nerve palsies. One patient presented with oculomotor palsy associated with abducens nerve palsy. In the case reported by Mikawa and colleagues, it is unclear whether the third nerve palsy is due to classic compression caused by uncal herniation or sagging of the brain due to SIH.

Spontaneous acute or chronic subdural haematomas found in patients on anticoagulation do not usually prompt additional investigation to identify the cause of the haemorrhage. This is the fourth case reported of SIH presenting with subdural haematomas in patients on anticoagulation. Schievink et al. reported three cases of elderly patients on a regimen of long-term anticoagulation when they developed chronic subdural haematomas. These three patients met the diagnostic criteria for SIH due to spontaneous spinal CSF leak.

The vast majority of cases of ophthalmoplegia caused by SIH resolve completely. In one case reported by Brady-McCreery and coworkers, a patient with bilateral trochlear nerve palsies and unilateral oculomotor nerve palsy due to SIH failed to resolve after standard treatment and required strabismus surgery.

SH can have a wide clinical presentation—from mild focal neurological deficits and/or headaches to coma. The presence of
painful third nerve palsy with a good history of postural headaches should prompt a diagnosis of SIH. Subdural haematomas in a patient with a normal level of consciousness are unlikely the cause of the oculomotor nerve palsy. Vascular pathology compressing the oculomotor nerve should be ruled out. The importance of a thorough history to better characterize headaches is highlighted by...
our case. The vast majority of oculomotor nerve palsies due to SIH resolve with standard treatment.

DISCLOSURES
Charlotte Dandurand and Charles Haw hereby declare that they have nothing to disclose.

STATEMENT OF AUTHORSHIP
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