An interesting case of headache
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Abstract

A 35-year-old businessman with a history of migraine with aura developed new neck pain while lying on a sofa in his home. He was given neck massage and physical therapy for a day, and subsequently after two days developed severe generalized headache when sitting or standing. He was hospitalized in his hometown. Investigations revealed venous sinus thrombosis and bilateral thin subdural collections for which he was anticoagulated. Headache improved and then worsened and became severe in all positions. He was then admitted under our care. MRI scan of the brain at our hospital showed left subdural hematoma with midline shift. It required urgent evacuation. His previous first brain MRI was re-evaluated. It showed characteristic features of spontaneous intracranial hypotension (SIH). If not recognized early, SIH results in various complications, some of which require immediate intervention. Any change in the pattern of headache in SIH one must alert the clinician due to the possibility of one of its complications.

Key Words

Headache, spontaneous intracranial hypotension, subdural hematoma, venous sinus thrombosis

Case Report

A 35-year-old businessman developed neck pain while lying on a sofa with his head propped up resting against the sofa edge in his home. He asked someone there to massage his neck. This was done vigorously. Almost immediately, the pain became severe and he was unable to turn his head. Next day he received neck physiotherapy. The pain persisted. Two days later, he saw a physician who prescribed analgesics and a cervical collar. For four days, the pain was unrelieved so he went to an orthopedic surgeon, who treated him symptomatically for eight days without improvement. He then developed vomiting and diarrhoea and the headache became generalized, worse when sitting or standing. He was hospitalized in his home town. MRI scan with venography showed thrombosis of the superior sagittal and sigmoid sinuses, and small bilateral subdural collections. He was treated with low molecular weight heparin, then oral anticoagulants and aspirin and was discharged. Two days later, he developed severe orthostatic headache and required readmission. MRI scan with venography showed no improvement. He was treated for a week again with low molecular weight heparin, and discharged on oral anticoagulants. He felt better for two days but the orthostatic headache recurred with vomiting. Over the next two days, the headache became continuous and unbearable.

On admission to our hospital, he had severe generalized headache in all positions, but no clinical signs. Incidentally the patient gave a 20-year-history of episodic bifrontal headaches preceded by a bright white linear visual aura.

MRI scan showed bilateral subdural collections [Figure 1d], the larger left one requiring evacuation on the same day after...
stopping anticoagulants and giving fresh frozen plasma and platelets. The postoperative period was uneventful. As the orthostatic nature of the initial headache suggested intracranial hypotension, previous imaging studies were reviewed. In the very first study, there were bilateral small subdural fluid collections [Figure 1a], downward descent of brainstem and cerebellum [Figure 1b], enlarged epidural venous plexuses at the craniocervical junction and an anterior, epidural CSF collection from the back of C2 vertebral body to the upper dorsal vertebrae [Figure 1c]. These features were consistent with intracranial hypotension due to CSF leak. Repeat venous MRI of the brain and screening of the whole spinal cord showed partial re-canalization of the previously thrombosed cerebral venous sinuses and only minimal bilateral subdural collections. The previously visualized spinal epidural collection remained. The patient declined to have any further invasive procedure. He opted for near total bed rest with adequate oral fluid intake. The thrombophilia profile was normal. At review one month later, he was symptom-free, apart from one episode of migraine with aura. CT scan of the brain showed a thin residual left subdural collection.

At his last follow-up two months later, he remained well. A repeat venous MRI of the brain and screen of the spinal cord showed no subdural hematomas, but the chronic intracranial venous sinus thrombosis and anterior epidural cervicodorsal fluid collection persisted.

Discussion

Clinically SIH is characterized by postural headache, which occurs or worsens while sitting or standing and is relieved by lying down. In young and middle-aged adults, it is an important cause of new onset headache that results from spinal CSF leak without any obvious trauma such as head injury or lumbar puncture. The associated and precipitating factors for these are weak dura at the site of tear, either as a part of a generalized connective tissue disorder, or due to trivial trauma to the spine such as bending, lifting or cervical spine manipulation. We feel that in our patient cervical spine massage might have aggravated the leak.

The Monro Kellie hypothesis states that when the skull is intact the total volume of brain, CSF and intracranial blood remains constant. As the volume of brain changes very little, decreased CSF volume as in SIH causes increased blood flow, mainly in the venous system and stasis, which may result in thrombosis. The subdural fluid collection also contributes to maintaining the intracranial volume. If the CSF leak continues, the brain sags, cortical veins rupture and a hematoma results.

![Figure 1(a-d): Image a, b & c are the very first MRI while image d is at the time of admission. (a) T2W coronal MRI brain showing bilateral thin subdural hygromas; (b) T2W transverse MRI cervical spine showing anterior epidural CSF fluid accumulation behind C2 and C3 vertebra (c) Contrast MR venogram brain showing very poor flow in superior sagittal sinus (d) MRI brain showing left subdural hematoma with midline shift](image-url)
Venous sinus thrombosis and subdural hematoma should be suspected when the pattern of headache changes from orthostatic to continuous as in this case.

The characteristic MRI features of SIH are presence of bilateral subdural hygromas, diffuse pachymeningeal enhancement, downward displacement of the brain at the tentorial hiatus and foramen magnum and dilatation of venous sinuses. One may also see brain stem herniation in secondary intracranial hypertension. This can mimic SIH as both produce headache and cranial nerve palsies. However, the orthostatic headache without papilloedema differentiates SIH from intracranial hypertension.

Conservative treatment for SIH is adequate hydration and bed rest. Sometimes it may resolve spontaneously, but the mainstay of treatment is epidural autologous blood patch, which is also an effective treatment for its complications. However, some complications may require conventional treatment: urgent evacuation of subdural hematoma and heparin for venous sinus thrombosis. Asymmetric bilateral subdural hematomas may not produce focal neurological deficits and may be missed. In addition, if venous sinus thrombosis is treated with anticoagulants without diagnosing the primary cause, complications may result.

The lesson from this case is that with orthostatic headache in any patient, one must exclude SIH by MRI. If SIH is complicated by venous sinus thrombosis and/or subdural haematoma, diagnosis will be more difficult because the initial orthostatic nature of the headache will have become continuous. In such cases, unless the underlying SIH has been identified, treatment will be unsuccessful.

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