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Spontaneous Intracranial Hypotension Following a Yoga Class: A Case Report

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Abstract Spontaneous intracranial hypotension (SIH) is an underdiagnosed syndrome and initially missed cause of headaches. The classical presentation is that of an orthostatic headache, mainly caused by spontaneous and difficult to detect spinal CSF leaks. The current case report describes a 38 year old female patient who presented with symptoms following a Yoga class. Despite being initially misdiagnosed, her clinical course and radiological findings made it clear that she had intracranial hypotension syndrome with bilateral chronic subdural hematomas and cerebellar descent. She responded to surgical drainage of both her subdural hematomas and conservative management for her suspected CSF leak. This case, to our knowledge, is the first report in the literature of spontaneous intracranial hypotension being caused by a Yoga class. The report highlights the fact that some cases of SIH can be difficult to diagnose as their symptoms may be very confusing. Thorough interrogation of patients is vital, as trivial efforts and some exercising activities could be the cause of spontaneous dural tears. Early recognition is important to prevent unnecessary investigations and procedures, and to minimize delay in treatment.

Keywords: *spontaneous intracranial hypotension, orthostatic headache, low pressure headache, CSF leak, Yoga*

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1. Introduction

Spontaneous intracranial hypotension (SIH) is an uncommon but increasingly recognized cause of headaches in young and middle-aged individuals mainly caused by spontaneous spinal cerebrospinal fluid (CSF) leaks. Initial misdiagnosis is rather common. Orthostatic headache is the main symptom and clinical examination is usually normal. Magnetic resonance imaging shows the characteristic association of: diffuse pachymeningeal gadolinium enhancement, venous dilatation, sagging brain and bilateral subdural collections. Despite various possible explanations, the pathogenesis of this clinical entity is still a motive of debate. Conservative treatment is initially attempted and spinal epidural blood patch is reserved for those patients who fail this approach. We describe the first reported case of Yoga associated with spontaneous intracranial hypotension.

2. Case Report

We present the interesting case of a 38-year-old Caucasian woman who was completely asymptomatic until finishing an intense Yoga class. She initially developed a diffuse and severe headache combined with cervical pain that was interpreted as a cervical muscle sprain. Given that her symptoms at the time were mainly in the neck, she was investigated with a cervical MRI that showed mild rectification with no other relevant radiological findings. She was then referred to physical therapy. As the following weeks evolved, she developed a classic orthostatic headache that improved with supine position and caffeine, and worsened when sitting or standing up. A brain MRI showed diffuse pachymeningeal gadolinium enhancement both surrounding the brain and cervical spine, with cerebellar tonsillar descent towards the foramen magnum, posterior fosa crowding, engorged cerebral venous sinuses, enlarged pituitary gland, decreased size of the ventricles and bilateral subdural fluid collections. A lumbar puncture was performed evidencing very low pressure and obtaining minimal amount of fluid. At this point, no diagnosis was reported by the treating physician and no treatment was recommended.

Her symptoms continued progressing and by the time she consulted our institution for the first time, three weeks later (six weeks after the onset of her symptoms), her postural headache had significantly worsen, nonetheless, her neurological examination was unremarkable. Upon reviewing her case, we felt her symptoms were typical and consistent with intracranial hypotension syndrome and her previous radiological findings confirmed our clinical

suspicion. A repeat brain and cervical MRI (Figure 1, Figure 2) showed that both subdural collections had significantly increased in size with flattening of her sulci over the convexity and her pachymeningeal enhancement also increased in size specially at the cervical region (Figure 3).

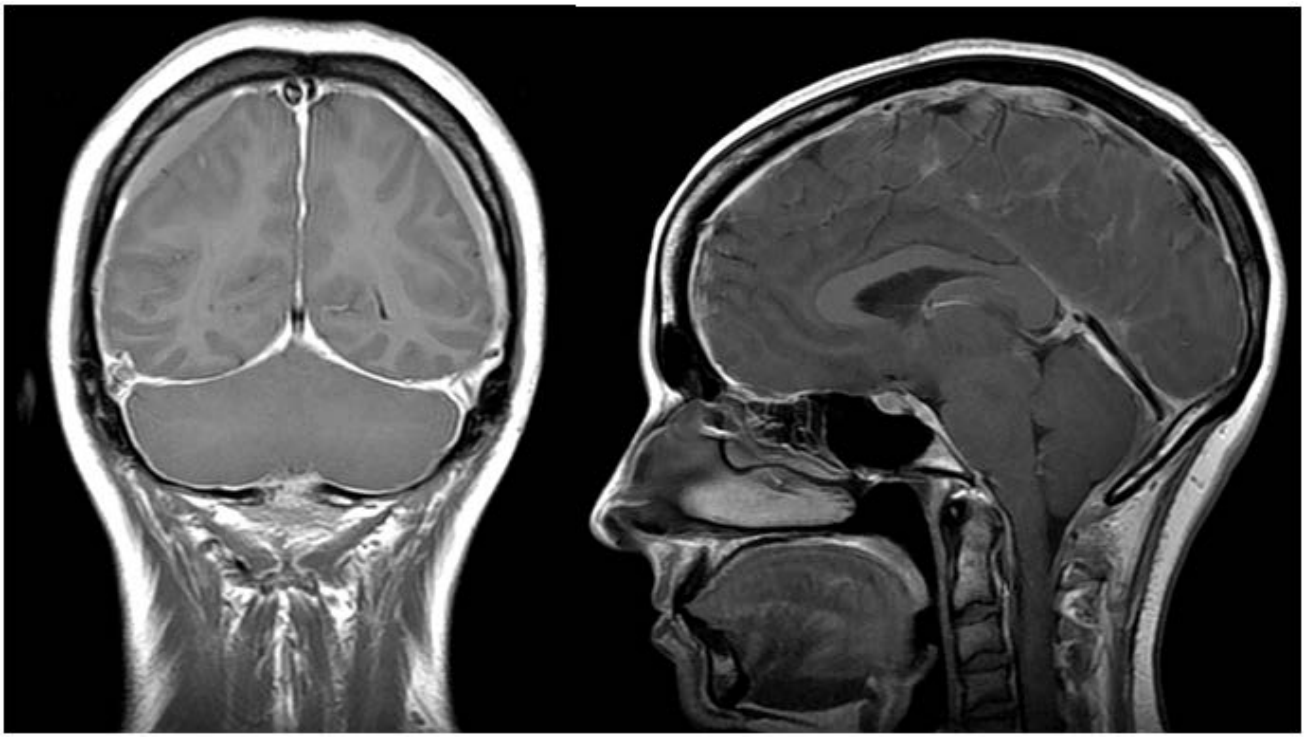

Figure 1. Gadolinium-enhanced T1 weighted MRI image demonstrating diffuse pachymeningeal enhancement and bilateral subdural fluid collections. a) Coronal view. b) Sagital image with mildly prominent pituitary gland and brain sagging

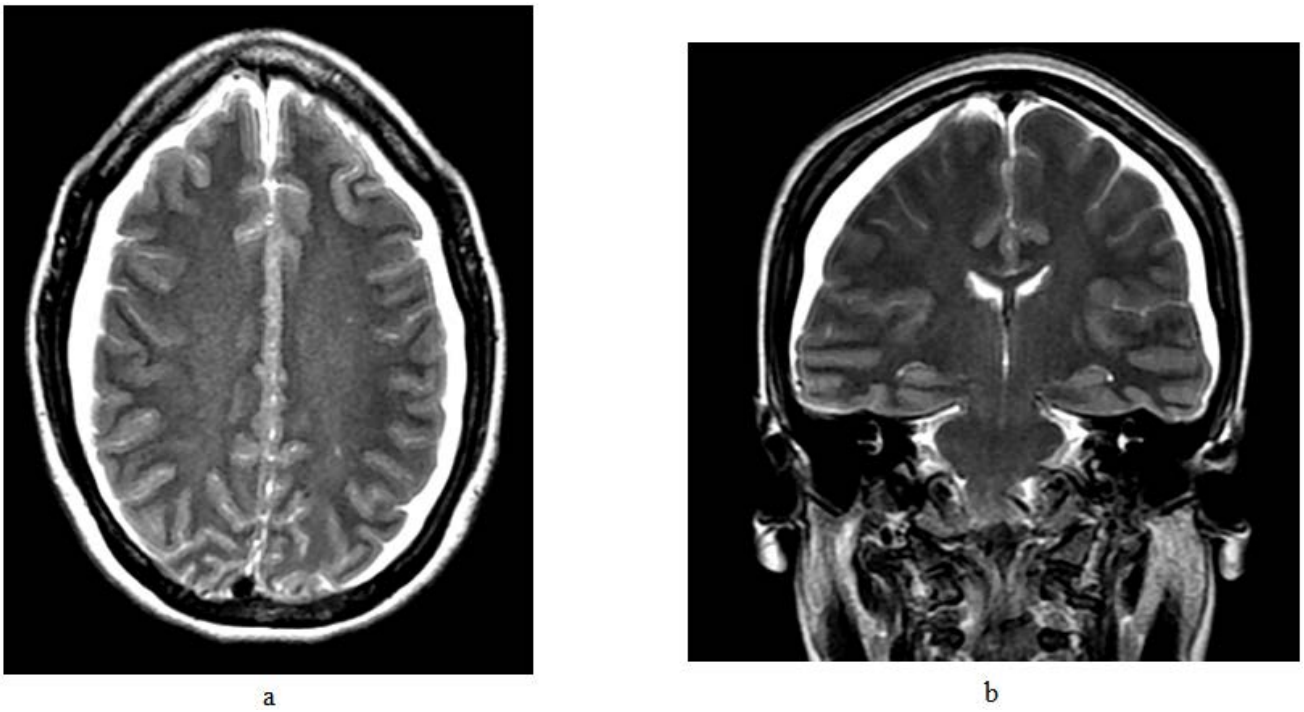

Figure 2. T2 weighted MRI of the brain showing bilateral fluid collections with flattening of both hemispheres. a) axial and b) coronal views

Given that her headaches had become almost unbearable and the collections had worsen, we decided to investigate the underlying cause and surgically treat them. She underwent a bilateral craniectomy and surgical drainage of her bilateral subdural collections (both under significant pressure). Her postoperative course was uneventful. A gadolinium Myelo MRI was performed trying to identify a possible CSF leak which was not demonstrated. Given that she never attempted a conservative approach with bed rest, she was discharged home under absolute bed rest and caffeine intake, with progressive tolerance to orthostatism. Her previous symptoms disappeared without further action needed and she gradually returned to her activities of daily living.

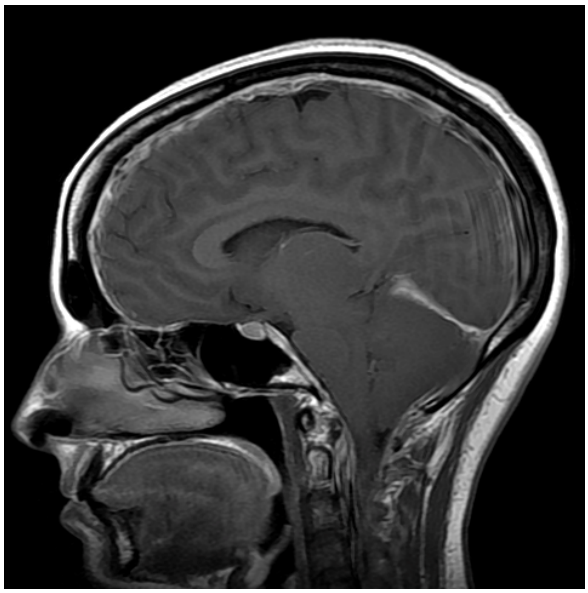

Figure 3. Gadolinium-enhanced T1 weighted sagital MRI image demonstrating brain sagging and posterior fossa crowding with diffuse pachymeningeal enhancement extending towards the cervical spine, being specially prominent and thick at the C2 level

3. Discussion

Intracranial Hypotension may be spontaneous (no evident cause found) or provoked by interventions such as a lumbar puncture, unintentional dural puncture during spinal anesthesia, or after neurosurgical procedures. Although it is generally accepted that Spontaneous Intracranial Hypotension (SIH) is due to a CSF leakage, the exact cause of the spontaneous leak remains unclear in many cases and its true incidence is therefore unknown [1,2,3,4]. When present, the leak most commonly arises from the spine, and less frequently from the skull base [1,2].

Some reports suggest that spontaneous spinal CSF leaks may be related to an underlying preexisting structural weakness of the dura rendering it more vulnerable to the effect of trivial trauma or unusual strains [1-6]. Reported triggering events include lifting small or large items, straining, stretching, coughing, positional changes, sporting activities, roller coaster rides, falls, and rarely, activities such as pilates or chiropractic manipulation [3,5,6,7,8]. Connective tissue disorders such as Marfan syndrome, Ehlers-Danlos syndrome type II, autosomal dominant polycystic kidney disease, and isolated joint hypermobility, have also been associated with SIH [1,2].

According to the *International Classification of Headache Disorders [9]*, a headache attributed to SIH is usually accompanied by low CSF pressure (<60 mm CSF) and/or has evidence of CSF leakage on imaging. Clinically, orthostatic (or postural) headache is the hallmark of this condition [1] typically reaching its highest intensity within a few seconds or minutes of being upright and decreasing upon lying down. It may be generalized or localized to the frontal, temporal or, most frequently, the occipital or suboccipital regions [1,6]. The severity is variable and correlates poorly with the degree of intracranial hypotension [3]. Associated symptoms may include neck pain or stiffness, nausea, vomiting, diplopia, blurred vision, tinnitus or altered hearing, facial numbness or pain [1,10,11].

There are several explanations to the headaches in SIH. The downward displacement of the brain due to loss of CSF buoyancy is the most plausible mechanism used to explain the typical headache in SIH. This forced shift causes traction on pain-sensitive structures, predominantly the dura, resulting in a headache provoked by orthostatism. The compensatory response of cerebral vasodilatation to the loss of CSF volume, evident as pachymeningeal enhancement on cranial MRI, may also contribute to pain. Alongside, this downward displacement of the brain may result in stretching of some cranial nerves; this mechanism may explain the other associated symptoms in SIH described previously [12].

Clinical presentation is often highly suggestive of the diagnosis, however, because of the nonspecific nature of the symptoms, combined with unfamiliarity with the condition, many cases probably remain undiagnosed [11]. The initial approach is a thorough clinical history of the patient´s symptoms with a high index of suspicion. Imaging is used to confirm the diagnosis and to try to localize a possible leak in order to plan treatment [1,3,11,12].

Lumbar puncture is usually not necessary for the diagnosis, nevertheless, low CSF opening pressure on spinal tap is one of the diagnostic criteria suggested by the *International Headache Society* [9].

MRI with gadolinium enhancement is the imaging modality of choice. The most common and typical radiological findings on MRI are: subdural fluid collections, enhancement of the meninges, engorgement of venous structures (sinuses), pituitary enlargement, and downward displacement ("sagging") of the brain. Subdural hematomas are fairly common and can be among the first findings of SIH [1,13,14,15,16]. Although MRI with gadolinium enhancement has become the first-line diagnostic radiological tool, it is not usually helpful in localizing the exact site of a CSF leak. CT myelography, radionuclide cisternography and MRI myelography with gadolinium have all been reported to be useful in detecting the site of CSF leak with varying degrees of success [6,7,16,17,18] and either one or in combination are

commonly used in cases of SIH. When localized, the CSF leak may appear as a subtle or as a diffuse extravasation of contrast. The majority of leaks are at the cervico-thoracic junction or thoracic spine. Occasionally, multiple simultaneous CSF leaks are demonstrated on myelography at different spinal levels.

Given that some of the imaging studies mentioned above may be somehow invasive, often fail to pinpoint the site of leakage [7], and in many cases do not alter our management plan, some authors believe that striving to search for a dural tear is essentially unnecessary [19].

Initial conservative management with bed rest, hydration (or, more accurately, overhydration), caffeine, and analgesia is generally the first course of action. Steroids, theophylline, abdominal binders or corsets have also been used with inconclusive evidence of efficacy [4,7,20].

Despite the subdural collections being a frequent component in these patients, the decision to surgically drain them as the mainstay of treatment is still controversial [13,14,21]. They seem to occur secondary to the tearing of small bridging veins caused by the downward traction on the brain as the CSF volume decreases and usually respond to conservative management. Nonetheless, patients with clinically important or enlarging subdural hematomas or progressive debilitating symptoms seem to have a more defined indication to proceed with surgery and may require neurosurgical intervention [12,20].

For those patients that fail to respond to conservative management, an epidural blood patch (EBP) is the mainstay of treatment [7,14,20,22,23,24,25]. This procedure involves injecting autologous blood into the epidural space, increasing the total volume within the spinal canal and thus raising the CSF pressure. Patients often refer immediate relief of their symptoms. The mechanism of action is believed to be twofold. The first is an immediate volume replacement, and the second is the sealing of the dural defect, which may be delayed from the first action [6]. If the location of the leak has been identified, the blood patch can be targeted accordingly at that spinal level. Otherwise, a “blind” injection into the lumbar spine is often successful [7,12,20,24], or even injecting at different sites has been done with favorable results [1,26,27]. It is worth mentioning at this point that the hypothesis involving intravenous hypotension as the pathogenesis in developing SIH is of particular importance. The loss of pressure inside the venous system may create a differential pressure gradient between venous and CSF compartments, possibly disrupting the dura at one or even several locations. Furthermore, this theory may also explain the success of unselective (or blind) epidural blood patches producing tamponage in the epidural space as well as compressing the leakage sites. [28].

Alternative treatments in special situations or when EBP fails include epidural saline infusions, epidural fibrin glue, fibrin glue and blood, and even epidural infusion of dextran (rarely practiced) [4,20]. However, the effectiveness of these measures has not been well established due to limited data. Surgical repair is a last resource treatment only when conservative treatment fails and the source of the leak has been clearly identified.

4. Conclusion

Spontaneous intracranial hypotension (SIH) is a common but unrecognized cause of headaches. Initial misdiagnosis is frequent and patients tend to suffer progressive and debilitating symptoms before a clear diagnosis is made. Early recognition and clinical awareness of spontaneous intracranial hypotension is therefore important to prevent unnecessary investigations and a delay in treatment. Yoga may be a risk factor for the development of a spontaneous cerebrospinal fluid leak in certain patients.

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