Cavernous Sinus Swelling and Abducens Nerve Paresis Due to Intracranial Hypotension

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Abstract:
The most common neurological symptom of spontaneous intracranial hypotension (SIH) is abducens nerve paresis, and the precise pathophysiology is unclear. The accepted explanation is traction on the cranial nerves caused by the downward displacement of the cranial content. We herein report magnetic resonance imaging of SIH that can explain the mechanism underlying abducens nerve paresis. The cavernous sinuses were particularly thickened compared with the surrounding dura. This phenomenon can be explained by venous swelling, which can occur after leakage of cerebrospinal fluid in a closed cavity. This swelling pushes the abducens nerve up, which then causes abducens nerve paresis.

Key words: spontaneous intracranial hypotension, hypertrophic pachymeningitis, abducens nerve paresis, cavernous sinus, digital subtraction angiography

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Introduction
Spontaneous intracranial hypotension (SIH) is a clinical syndrome in which a decreased cerebrospinal fluid (CSF) volume results in headache, nausea, vomiting, or double vision. The most common neurological symptom of SIH is abducens nerve paresis (1). It also features diffuse pachymeningeal gadolinium enhancement on magnetic resonance imaging (MRI) and is difficult to distinguish from hypertrophic pachymeningitis (HP) (2).

We herein report a new MRI finding of SIH that can explain the mechanism underlying abducens nerve paresis.

Case Report
A 36-year-old woman presented with headache, double vision, and nausea. She had no history of trauma. Headache was reduced in the supine position and worsened in the sitting position. Her consciousness was clear, and she had no fever or neck rigidity. Right abducens nerve paresis was observed at admission, and new left-side paresis appeared after hospitalization. Inflammatory conditions, such as rheumatoid arthritis, anti-neutrophil cytoplasmic antibody (ANCA)-related diseases, IgG4-related diseases and sarcoidosis, were negative on a laboratory examination. Malignant tumors and infections were also excluded.

Cerebrospinal puncture in the side-lying position was successful, but her CSF could not be collected due to a low CSF pressure. Cerebrospinal puncture in the sitting position showed a low initial pressure (10 mmH₂O). MRI showed diffuse enhancing meningeal thickening and a high intensity on T2-weighted imaging. In addition, the pituitary gland was enlarged. In particular, the bilateral cavernous sinuses (CS) were swollen (Fig. 1A).

HP was suspected, and a biopsy was considered. Spinal MRI revealed a floating dural sac sign and C1-2 false-localizing sign (Fig. 1B). Digital subtraction angiography (DSA) showed enlarged cortical and medullary veins (Fig. 1C). A diagnosis of SIH was established based on these findings (3-5).

With conservative management, her symptoms improved within a week. Magnetic resonance myelography and radioisotope cisternography (111-I DTPA) performed afterward did not reveal any leakage points, although the examination may have been performed too late because the leaking...
Figure 1. (A) From the left, axial view of T2-weighted imaging (T2WI), and coronal and sagittal view of enhanced T1-weighted imaging (T1WI). Magnetic resonance imaging showing diffuse and enhanced meningeal thickening with a high intensity on T2WI. In particular, the bilateral cavernous sinuses (CSs) were swollen compared with the surrounding dura. (B) The left, axial and sagittal views of T2WI and sagittal view of T1WI. Spinal MRI revealed a floating dural sac sign and C1-2 false-localizing sign. (C) Before treatment, digital subtraction angiography revealed dilatation of the peripheral veins of the brain. (D) Coronal and sagittal view of enhanced T1WI. After the patient’s symptoms improved, the swelling and enhancement disappeared, a dramatic finding at the CS site.

points were believed to have closed by then. Her headache and oculomotor nerve palsy improved, and MRI showed that the dural thickening and contrast effects were attenuated, while the CS and pituitary gland had returned to normal size (Fig. 1D). The abducens nerve is the only nerve located in the CS (Fig. 2A), so a CS structural change may have been the cause of the abducens paresis.

Discussion

Dura thickening and enhanced findings on MRI are characteristic of SIH and closely resemble those of HP. A dural biopsy-based report of a patient with SIH showed that the condition was not an inflammatory reaction, in contrast with HP (6). The SIH findings on MRI are caused by venous engorgement, not the dura mater. It appears as venous dilatation on DSA (7). The present case showed enlarged cortical and medullary veins (Fig. 1B). Furthermore, many case reports have pointed out that the Valsalva maneuver exacerbates the symptoms of SIH, suggesting that the condition is associated with the intracranial venous system.

The CS swelling can be explained by the Monro-Kellie
hypothesis. While the intracranial space is constant, a decrease in intracranial pressure caused by the loss of CSF volume leads to the extension of the soft venous wall to fill the free space. In the current case, the CS was particularly thickened compared with the surrounding dura. This phenomenon cannot be explained without venous swelling and is a unique feature of SIH. A biopsy is required for the diagnosis of HP; however, this procedure can be lethal in cases of SIH. Therefore, ruling out SIH is important, and this feature can help distinguish SIH from HP.

The precise pathophysiology underlying the abducens paresis observed in SIH is not clear. The most accepted explanation is traction on the cranial nerves caused by the downward displacement of the cranial content (8). The abducens nerve has a long course and is likely to be susceptible to the traction-related injury that occurs when the brain is depressed; this finding is subjective (9). In patients with SIH, thickening of the surrounding venous structures can lead to narrowing of the intracranial space, especially the brain stem; thus, it appears depressed.

Why abducens neuropathy does not occur after craniotomy, during which the brain moves downward because of a reduction in the CSF and the entry of air into the skull, remains unclear. The abducens nerve passes through Dorello’s canal, below Gruber’s ligament, and reaches the CS (Fig. 2B) (10). The CS is surrounded by bones anteriorly, medially, and inferiorly, and it can swell only upward and posteriorly in the setting of SIH. Therefore, CS swelling pushes the abducens nerve up, as this nerve is sandwiched between these structures, and can cause neuropathy. Craniotomy does not cause abducens neuropathy, as CS swelling can only occur after leakage of CSF into a closed cavity, as described above. Given that patients with abducens nerve paresis commonly exhibit a carotid cavernous fistula and CS dural arteriovenous fistula, which increase the pressure inside the CS, the abducens neuropathy observed in SIH can also be explained by CS swelling.

Several limitations associated with the present study warrant mention. First, we cannot demonstrate abducens neuropathy on an autopsy because it develops under the special conditions described above. Second, the abducens neurons, including both Dorello’s canal and Gruber’s ligament, are minute structures which are not visible on MRI. To address these issues in greater detail, the development of an image-inspection instrument and the accumulation of cases are needed.

The authors state that they have no Conflict of Interest (COI).

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