Upward gaze palsy and convergence insufficiency as a rare presentation of primary intraventricular haemorrhage

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SUMMARY

A primary intraventricular haemorrhage (PIVH) usually presents with non-localised neurological symptoms since the haematoma is limited to the ventricles. However, it is sometimes associated with focal neurological signs, whose pathophysiologies are not confirmed. Here, we report on a case of PIVH who showed rare manifestations in the acute stage: upward gaze palsy and convergence insufficiency. The CT and MRI showed intraventricular haematoma without evidence of parenchymal haemorrhage, local mass effect around midbrain or hydrocephalus. There had been bilateral papilloedema, and it resolved along with improvement of the ophthalmic symptoms, suggesting a possible causal relation to increased intracranial pressure. The ophthalmic abnormalities suggested injury of the rostral part of the midbrain, especially the region around the dorsal midbrain tectum. It should be known that PIVH is one of the causes of acutely developing upward gaze palsy and convergence insufficiency.

BACKGROUND

Intraventricular haemorrhage (IVH) is a severe form of intracerebral haemorrhage, which is frequently associated with serious neurological conditions due to brain tissue injury and hydrocephalus.[1 2] Primary intraventricular haemorrhage (PIVH) is a rare form of IVH, which is not associated with discernable parenchymal causes, comprising only 3% of IVH in adults.[3] PIVH usually presents with non-localised symptoms, headache, vomiting and altered mental status,[4 5] while it is rarely associated with focal neurological deficits.[6] We report on a case of PIVH who showed rare manifestations, upward gaze palsy and convergence insufficiency in the acute stage, and we discuss the pathophysiology of the neurological deficits.

CASE PRESENTATION

A 64-year-old man was transported to our emergency department because of worsened headache and vomiting that had suddenly developed the day before and was associated with diplopia in all eye positions. He had a history of untreated hypertension for several years. On admission, he was alert, and his vital signs showed high blood pressure (190/116 mm Hg) with a normal heart rate (72 bpm). The neurological examination revealed nuchal rigidity as well as conjugated upward gaze palsy and convergence insufficiency (figure 1A). There were no other neurological abnormalities. Ocular fundus findings showed bilateral papilloedema (figure 2B), suggesting increased intracranial pressure.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis included meningitis or intracranial haemorrhage such as subarachnoid haemorrhage, intracerebral haemorrhage or IVH. IVH usually develops secondary to subarachnoid haemorrhage, parenchymal haemorrhage or trauma. It also can be a result from hypertension, vascular malformations, intraventricular tumours and other rare causes (moyamoya disease, cerebral amyloid angiopathy or cerebral venous thrombosis).

INVESTIGATIONS

Blood tests revealed no remarkable abnormalities. An ECG showed left axis deviation and T wave inversion of V3–V5 leads but did not meet the voltage criteria of left ventricular hypertrophy. A chest X-ray showed no remarkable cardiomegaly. CT (figure 2A) and MRI (figure 3) taken on admission showed IVH dominant in the left lateral ventricle, but no evidence of parenchymal lesion and subarachnoid haemorrhage. MRI (figure 3) and MR angiography showed no aneurysms, vascular malformations, nidus of vessels, intraventricular tumours, microbleeds on T2-weighted gradient-recalled-echo imaging or flow voids in the dural sinuses.

TREATMENT

Because the possible causes could be excluded, the patient was diagnosed as having PIVH. As he was alert, external ventricular drainage or

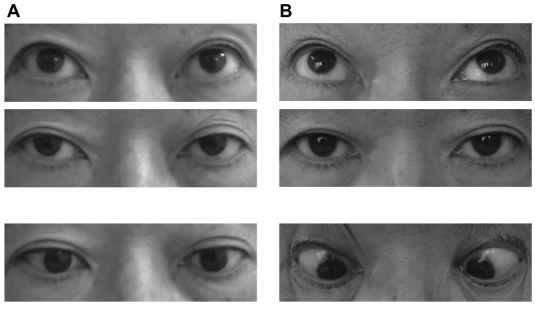

Figure 1 Photographs of eye movements. Top, upper gaze; middle, primary position; bottom, convergence. (A) Upward gaze palsy and convergence insufficiency were observed on admission. (B) Eye movements normalised 4 weeks after onset.

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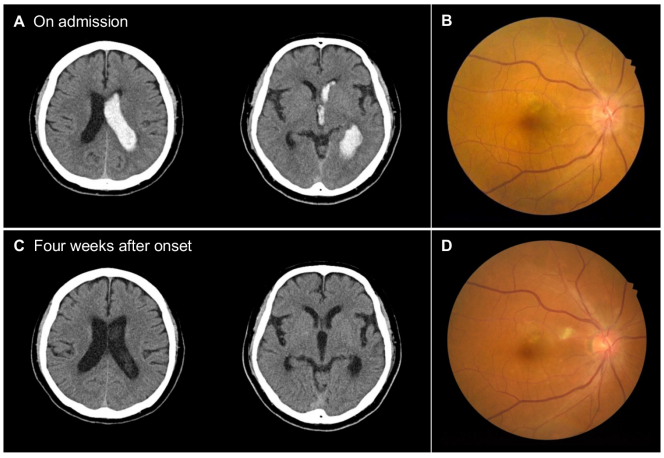

Figure 2 The CT imaging and ocular fundus findings of the right eye on admission (A, B) and 4 weeks after admission (C, D). (A) Intraventricular haematoma dominated in the left lateral ventricle on admission. (B) The papilloedema of the right fundus, which was also present on the left eye (photograph not shown). (C) A mostly absorbed intraventricular haematoma in the fourth week after admission. (D) The papilloedema was improved, which was also observed on the left eye (photograph not shown).

intraventricular thrombolysis were not carried out. Conservative treatment, including head-end elevation and blood pressure management was initiated under intensive neurological care.

OUTCOME AND FOLLOW-UP

Three weeks after onset, the headache and the diplopia improved, and the upward gaze palsy or the convergence insufficiency were resolved (figure 1B). Four weeks after onset, the follow-up head CT scan showed a mostly absorbed haematoma in the ventricle (figure 2C). The fundus findings showed an improvement of the papilloedema (figure 2D). Five weeks after onset, he became free of the symptoms and was discharged.

DISCUSSION

Our case of PIVH showed localised neurological deficits: upward gaze palsy and convergence insufficiency. While PIVH is rarely associated with focal neurological signs,[6] it can present with cranial nerve palsy, hemiparesis or ataxia.[3 5–14] Oculomotor abnormalities, including impaired upward gaze,[14] have also been reported, though most of the literature has not reported the details.[3 10–14] Accordingly, the underlying pathophysiology of the oculomotor abnormalities has not yet been clarified.

The simultaneous abnormalities of conjugated upward gaze and convergence suggested a specific localisation of brain injury in the dorsal midbrain tectum or its nearby structures in the midbrain. The main structures involved in the vertical gaze are the rostral interstitial nucleus of the medial longitudinal fasciculus (riMLF), the interstitial nucleus of Cajal (INC) and the posterior commissure (PC).[15 16] The PC is composed of axonal fibres of neurons including the INC. While elevator muscles (superior rectus and inferior oblique) are innervated by riMLF, INC and PC, depressor muscles (inferior rectus and superior oblique) are innervated by riMLF without the involvement of INC or PC.[15] Because of these differences in neural circuits, isolated upward gaze palsy suggests injury of the PC or INC.[16] Interestingly, the main structures involved in the convergence are also thought to be located in the midbrain reticular formation, as illustrated by Dr Mays in 1984.[17 18] Even if the loss of convergence could arise from bilateral trochlear nerve dysfunction, the trochlear nerve crosses the midline in the posterior tectum near the posterior commissure. Based on these anatomical insights, the site responsible for the oculomotor symptoms in our case is estimated to be located in the midbrain, especially in the dorsal midbrain tectum.

As for the brain injury mechanism in PIVH, four possible scenarios have been suggested: increased intracranial pressure (ICP), haematoma-exerted local mass effect, blocking of the cerebrospinal fluid (CSF) flow and the haematoma-derived toxicity.[2] In our case, there was transient papilloedema, which improved along with the resolution of the symptoms, indicating the causal relationship of increased ICP with brain injury. Indeed, increased ICP is known to cause a 'false localising sign',[19] and upward gaze palsy has been reported as one of these signs.[20 21] Other possible mechanisms are not likely, or there is neither supportive nor conflictive evidence of the contribution in our case.

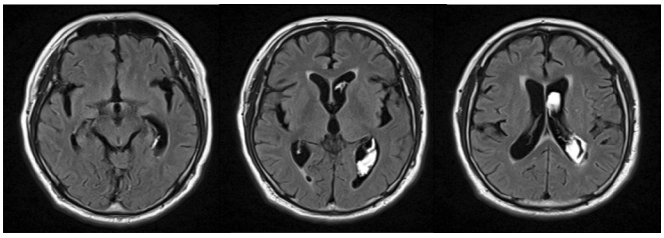

Figure 3 The fluid-attenuated inversion recovery MRI taken on admission. There was no vascular abnormality in the ventricle wall and no parenchymal lesion in the midbrain.

Learning points

- It is important to know that upward gaze palsy and convergence insufficiency can occur as the initial symptoms of primary intraventricular haemorrhage.
- The ophthalmic abnormalities suggested injury of the rostral part of the midbrain, especially the region around the dorsal midbrain tectum.
- Increased intracranial pressure could be a potential mechanism of midbrain injury in our case.

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