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CASE REPORT

Rare valiant vertical one-and-a-half syndrome without ipsilateral upward gaze palsy in a patient with thalamomesencephalic stroke

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Abstract

Bilateral upward and ipsilateral downward gaze palsy due to a unilateral thalamomesencephalic stroke is called vertical one-and-a-half syndrome (VOHS). Here, we report a valiant VOHS case who presented contralateral upward and ipsilateral downward gaze palsy due to a unilateral thalamomesencephalic stroke. The neuronal fiber connections associated with vertical gaze are not completely understood, so the present case provides an important proof to obtain a better understanding of vertical gaze mechanisms.

Introduction

Bilateral vertical gaze palsy was previously reported as a result of a unilateral stroke of the rostral midbrain.[1,2] Here, we report a valiant vertical one-and-a-half syndrome (VOHS) case who presented contralateral upward and ipsilateral downward gaze palsy due to a unilateral thalamomesencephalic stroke.

Case report

A 62-year-old man became aware of diplopia and ataxia when he woke up one morning and was unable to walk long without seeking the support of a wall. The next morning, he visited our clinic, when he showed left eyelid ptosis with the left eye directed 10° upwards (Fig. 1a), the right eye displaying upward gaze palsy, and mild downward gaze palsy of his left eye (Fig. 1b). This vertical gaze disorder involved both voluntary saccades and smooth pursuit. Convergence was absent (Fig. 1b). Vertical optokinetic nystagmus and vertical oculocephalic response were also impaired. These eye symptoms were similar to VOHS that causes bilateral upward gaze palsy and unilateral downward gaze palsy, although his left eye showed a full upward gaze (Fig. 1b, top center). He walked unsteadily with truncal ataxia. Brain MRI revealed a small high-intensity area on a diffusion-weighted image (DWI) in the left midbrain-thalamus transition area (Fig. 1c–e, arrows). Although he received chemotherapy for lung adenocarcinoma 1 year earlier, gadolinium (Gd)-enhanced MRI did not show any evident metastatic brain tumor (Fig. 1f). MR angiography did not clearly show stenosis of the basilar and left posterior cerebral artery (PCA) where the medial midbrain-feeding artery (paramedian artery) diverged (Fig. 1g, an arrow).

Upon admission to our hospital, his blood pressure was mildly high (151/88 mmHg). Laboratory data indicated an elevated erythrocyte sedimentation rate (ESR, 62/101 mm), CRP (1.59 mg/dL), total cholesterol (303 mg/dL), LDL cholesterol (221 mg/dL), d-dimer (1.5 mg/dL), and tumor markers (CEA 30.70 ng/mL and CYFRA 3.6 ng/mL). An ultrasound of the carotid artery showed atherosclerotic plaques in bilateral carotid bifurcations. Based on the diagnosis of an acute ischemic stroke, he began to receive intravenous edaravone and argatroban with oral administration of aspirin and clopidogrel. The disturbed eye movement improved completely until the 7th hospital day, so he was discharged from our hospital to a rehabilitation hospital for successive rehabilitation of truncal ataxia.

Discussion

The present case presents such a rare case of eye movement disturbance consisting of a contralateral upward gaze and ipsilateral downward gaze palsy due to a unilateral thalamomesencephalic stroke.

Although the neuronal fiber connections associated with a vertical gaze are not been completely understood, bilateral vertical gaze palsy was previously reported as a result

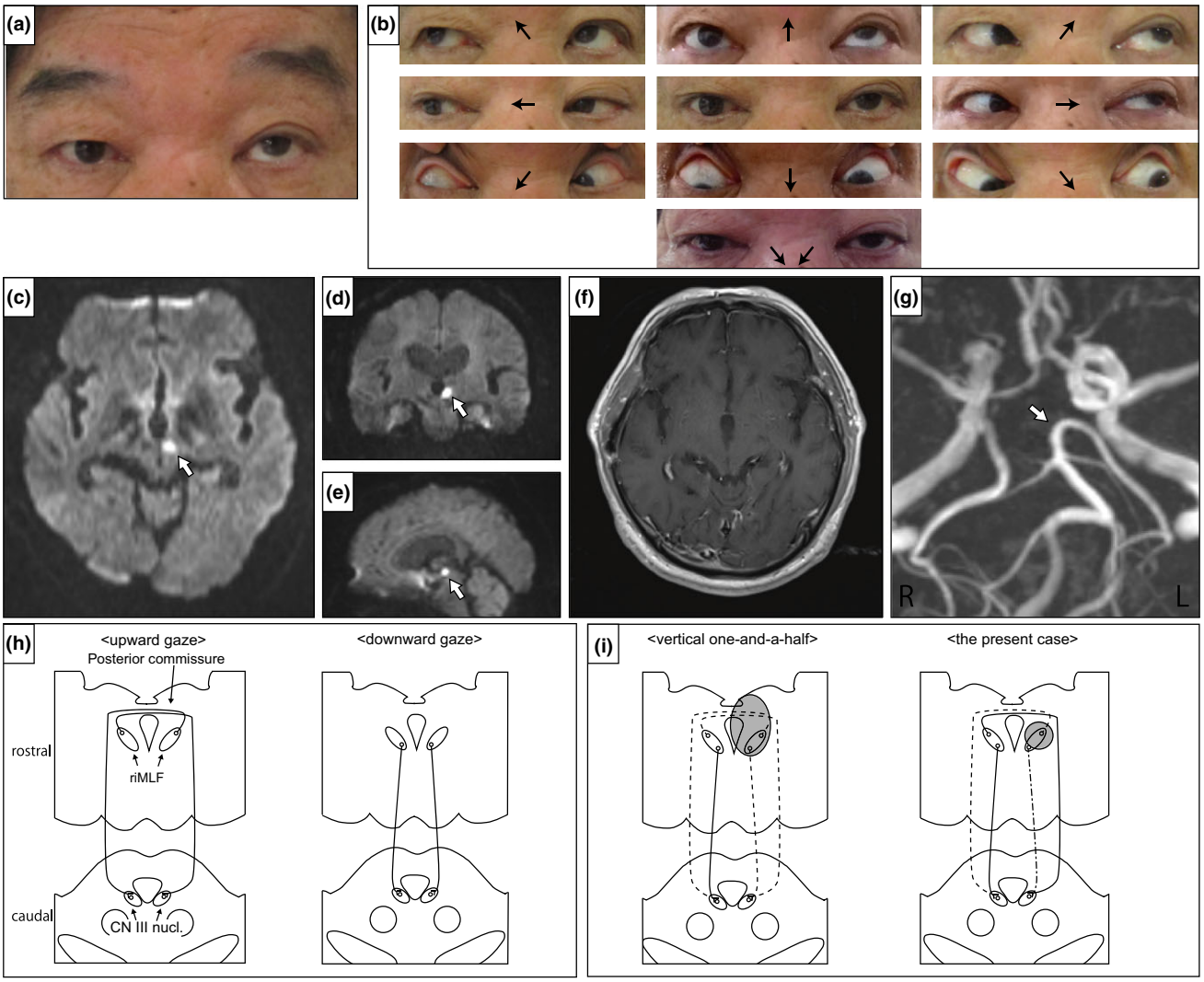

Figure 1 The left eye assumes a 10°position upwards in a straight ahead gaze (a) and mild downward gaze palsy (b), while the right eye shows evident upward gaze palsy (b). Convergence was absent (b). DWI images of a brain MRI, showing a small thalamomesencephalic stroke (c–e, arrows) without Gd enhancement (f). Brain MRA showing no evident stenosis of the basilar artery or left PCA (g, an arrow). Hypothesis of vertical eye movement (h): riMLF controls contralateral upward and ipsilateral downward gaze. Classical VOHS (i, left) involves bilateral upward and ipsilateral downward gaze (dotted lines) by a thalamomesencephalic lesion (left eclipse). The present case (i, right) involved only contralateral upward (dotted lines) and ipsilateral downward gaze (chain lines) due to a small thalamomesencephalic stroke (eclipse).

of a unilateral stroke of the rostral midbrain.[1,2] In that situation, upward eye movement is affected by fibers from the dorsal lateral part of the contralateral rostral interstitial nucleus of medial longitudinal fasciculus (riMLF) through the posterior commissure, and downward eye movement is affected by fibers, mainly from the medial caudal part of the ipsilateral riMLF (Fig. 1h). Thus, a unilateral thalamomesencephalic stroke covering both the riMLF and posterior commissure causes bilateral upward gaze and ipsilateral downward gaze palsy (i.e. classical VOHS)[3,4] (Fig. 1i, left). On the other hand, there is only one report without ipsilateral upward gaze limitation,[5] as in our present case, in which the stroke lesion was estimated to be smaller than that of past cases and limited to the dorsal lateral part of the riMLF (Fig. 1i, right). In the past[5] and in the present case, the efferent fiber from the left dorsal lateral riMLF associated with contralateral upward gaze, as well as a part of the efferent fiber from the medial caudal riMLF associated with the ipsilateral downward gaze, is affected by a stroke (Fig. 1i, right, dotted lines).

In summary, the present case is a very rare valiant type of valiant VOHS and provides important clues to better understanding vertical gaze mechanisms.

Acknowledgment

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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