



Case Report

Midbrain hemorrhage presenting with isolated downward gaze palsy: A case report

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ABSTRACT

Background: Diseases presenting with only downward gaze palsy are extremely rare particular in cerebral hemorrhage.

Case Description: A 63-year-old man with no medical history developed a downward-dominant vertical gaze paralysis with a convergence disorder. Computed tomography and magnetic resonance imaging showed a small hemorrhage of 13 mm in diameter in the right midbrain tegmental area. The patient was conservatively treated. His symptoms showed a gradual improvement from upward gaze paralysis and convergence disorder followed by downward gaze paralysis. All symptoms disappeared in 3 weeks after the onset. The center of vertical eye movement was thought to be the rostral interstitial nucleus of medial longitudinal fasciculus (riMLF) and posterior commissure (PC).

Conclusion: In this case, bilateral riMLF and PC were impaired, resulting in bilateral vertical ocular motility disorder with upward gaze paralysis. Brainstem hemorrhage rarely presents with vertical gaze palsy. Most are cerebral infarctions, and few are due to cerebral hemorrhage. This case was important for better understanding the pathophysiology of an ocular motility disorder. We also summarized the characteristics of isolated vertical gaze palsy caused by cerebral hemorrhage.

Keywords: Downward gaze palsy, Midbrain hemorrhage, Posterior commissure, Rostral interstitial nucleus of medial longitudinal fasciculus (riMLF)

INTRODUCTION

Diseases presenting with vertical gaze palsy are rare and occur with vascular disorders or tumors in the dorsal midbrain.^[1] Parinaud syndrome and other pathological conditions presenting with upward gaze palsy are sometimes encountered, but only downward gaze palsy is rare. In particular, cerebral hemorrhage with such symptoms is extremely rare. We experienced a case of cerebral hemorrhage and vertical gaze palsy with downward gaze disturbance as the main symptom. In addition to considering the mechanism of its expression, we will examine the characteristics of vertical gaze palsy caused by cerebral hemorrhage.

CASE DESCRIPTION

A 63-year-old man referred to our hospital with a sudden onset of generalized headache and diplopia that worsened in all directions, especially looking downward. On admission, there were

no abnormalities in vital signs and blood tests. Neurological findings showed clear consciousness. Vertical gaze palsy, convergence disorder, bilateral light reflex loss, and diplopia, which worsened with downward gaze, were observed. Horizontal eye movements were maintained [Figure 1]. A head computed tomography (CT) scan on admission showed a $10 \times 13 \times 13$ mm hemorrhage in the tegmentum of the midbrain [Figure 2]. A head magnetic resonance (MR) imaging on the same day showed hemorrhage in the midbrain tegmentum and edema in the posterior commissure (PC) [Figures 3a and b]. Contrast-enhanced MR imaging and MR angiography showed no opacities suggestive of bleeding sources such as neoplastic lesions or abnormal vascular networks. Conservative treatment was performed by the diagnosis of mesencephalic tegmental hemorrhage. The CT scan the next day showed no expansion of hemorrhage, and from the 4th day onward, the symptoms of upper gaze paralysis tended to improve, predominantly on the left side. On the 5th day, diplopia and light reflex improved, and on the 6th day, the left-dominant downward gaze paralysis also improved. On the 12th hospital day, all ocular symptoms disappeared except for downward gaze paralysis, and MR imaging during the same period showed that the hematoma was disappearing and the edematous changes near the PC disappeared [Figures 3c and d]. On the 25th day, the symptoms of bilateral downward gaze paralysis disappeared, and the patient was discharged. T2*-weighted image on MR imaging after about 2 years showed residual hypointense region, indicating that cavernous malformation is the etiology of the hemorrhage. We planned to perform periodic imaging follow-up.

DISCUSSION

This is an extremely rare case of vertical gaze palsy presenting with isolated downward gaze palsy due to cerebral hemorrhage. We considered the mechanism.

The center of vertical eye movement is said to be associated with the rostral interstitial nucleus of medial longitudinal

fasciculus (riMLF), the Cajal nucleus, the PC, and the Darkschewitsch nucleus.^[3,6] The functions of the Cajal nucleus and the Darkschewitsch nucleus have not yet been elucidated. Based on animal experiments using monkeys, Buttner-Ennever *et al.* suggested that the riMLF, which is present in the endoprosthesis of the mesencephalic red nucleus, is the center of vertical gaze.^[5] In addition, commands from the burst neuron in the riMLF are transmitted mainly through the ipsilateral oculomotor nucleus and trochlear nucleus to the muscles of the eye, causing upward and downward movements.^[6] The nerve fibers for upward and downward gaze are different, and the supranuclear fiber bundles responsible for upward gaze emerge from the medial or dorsolateral part of the riMLF, cross at the PC, branch left and right, and input to each nucleus. The mainstream theory is that the supranuclear fiber bundles of downward gaze enter the nucleus without crossing from the riMLF.^[1,2,4,7,10] On the other hand, there have been sporadic cases of bilateral vertical eye movement disorders due to unilateral midbrain lesions,^[4,8,9,13] suggesting the possibility that some of the nerve fibers involved in downward gaze are crossed on the contralateral side. In other words, upward gaze disorders occur with bilateral riMLF disorders, PC disorders, unilateral riMLF disorders, and disorders after the crossing of the connecting fibers to the contralateral side at the PC. Defective downward gaze may occur when the defect is bilaterally confined to the riMLF or when the fibers near the origin of the fascicles are bilaterally affected.^[8]

To satisfy these conditions, the lesion must be extremely localized toward the midline, so it is extremely rare for downward gaze paralysis to appear alone. In this case, the main locus of hemorrhage was in the right midbrain tegmentum, right riMLF, and edematous changes extended to the PC and contralateral riMLF. In the downward direction, it was thought that the edematous change partially extended to the vicinity of the dorsolateral oculomotor nucleus, which is considered to be the center of convergence. It is presumed that the upper gaze palsy

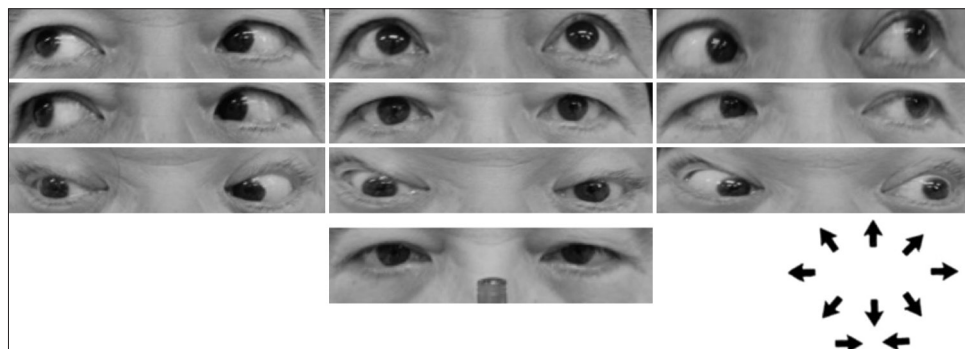


Figure 1: Eye movement test on admission. Vertical gaze palsy (upward and downward) was recognized. Each arrow indicates the direction of the eye's gaze in each photo.

was associated with the PC, and the lower gaze palsy was associated with dysfunction associated with bilateral riMLF and dorsolateral oculomotor nuclei due to hemorrhage and edematous changes [Figure 4]. On MR imaging, the edema of the PC improved at an early stage, and the function of the PC improved at that time, leading to the improvement of upper gaze paralysis. It was speculated that the symptoms improved in the order of downward gaze as the hematoma disappeared. Imaging also suggested a relationship between the symptoms and the site of injury [Figures 3 and 4]. The diplopia was thought to be associated with downward gaze disturbance and disappeared when the symptoms of downward gaze paralysis improved.

Most of the cerebrovascular diseases that cause vertical gaze palsy are caused by cerebral infarction, and cerebral hemorrhage has so far occurred in only six cases, including this case [Table 1].^[8,11-13] In all cases, small hemorrhages with about 1 cm diameter were localized in the midbrain tegmentum. The male-to-female ratio was 4:2, and the average age was 52.3 years old. The cause of bleeding was mostly unknown, and only one patient had a history of hypertension. Bleeding was on the right side in all cases. Although all cases showed upward

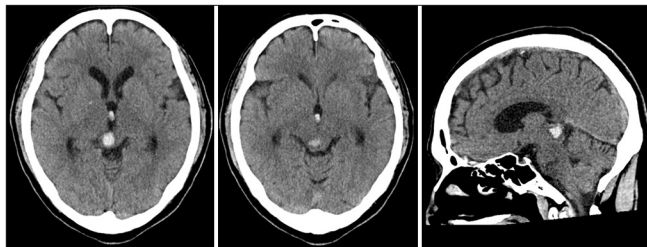


Figure 2: CT on admission. Hemorrhage of 10 × 13 × 13 mm in diameter in the right midbrain tegmental area. The hematoma was predominantly on the right side and partially contralateral.

gaze paralysis, half of the cases also showed downward gaze paralysis. None of the cases exhibited only downward gaze. Disturbance of consciousness often occurs in lesions of the tegmental part of the midbrain, and in such cases, it is difficult to evaluate eye movement disorders. Since lesions that are large enough not to disturb consciousness and that cause bilateral

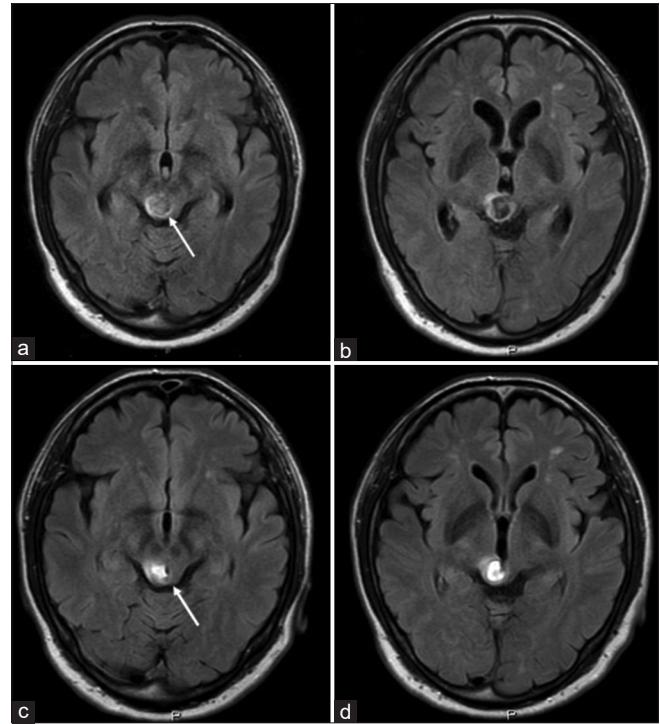


Figure 3: Time course of magnetic resonance imaging ([a and b] on admission [c and d] after 11 days). (a and c) are slice of the level of pineal gland, and (b and d) are slice of the upper level of pineal gland. The hematoma remained in the right cover and the edematous change of the posterior commissure disappeared (arrow).

Table 1: Summary of cases with vertical gaze palsy caused by intracerebral hemorrhage.

Case	Author	Year	Location	Lesion	Size (mm)	Vertical gaze palsy	Interval to recovery (weeks)	Prognosis
1	Sand <i>et al.</i> ^[11]	1986	Tectum, Tegmentum	Right riMLF, PC	15	Upward	12	MD
2			Tectum, Tegmentum, Crus cerebri	Bilateral riMLF, PC	25	Upward/Downward	NA	NA
3	Uehara <i>et al.</i> ^[13]	1990	Tectum, Tegmentum,	Right riMLF, PC	20	Upward	4	MD
4	Kawabata <i>et al.</i> ^[8]	1996	Tegmentum	Right riMLF, PC	13	Upward/Downward	12	MD
5	Takeuchi <i>et al.</i> ^[12]	2005	Tegmentum	Right riMLF, PC	13	Upward	1	GR
6	Present case	2020	Tectum, Tegmentum	Bilateral riMLF, PC	13	Upward/Downward	3	GR

GR: Good recovery, MD: Moderate disability, NA: Not applicable, PC: Posterior commissure, riMLF: Rostral interstitial nucleus of medial longitudinal fasciculus

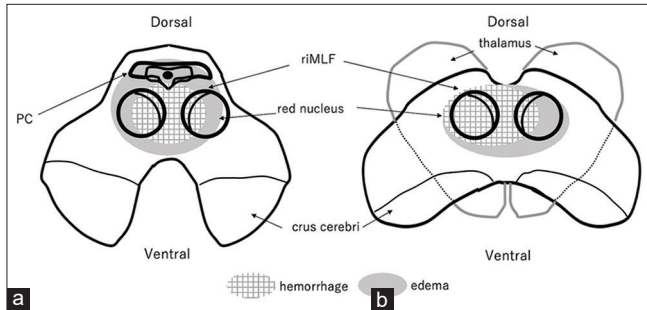


Figure 4: Schema of hemorrhage site. (a) is the level of pineal gland, and (b) is the upper level of pineal. In this case, bilateral rostral interstitial nucleus of medial longitudinal fasciculus had hematoma and posterior commissure had edematous changes.

riMLF impairment are extremely localized, downward gaze paralysis is thought to be rare. As for the prognosis, complete or partial improvement was observed in more than half of the patients, and most of them improved within 1 month of onset. It seems that the bleeding was small and the degree of parenchymal injury and surrounding edema was mild.

CONCLUSION

We report a case of cerebral hemorrhage in the midbrain tegmentum presenting with isolated downward gaze paralysis. Anatomically, the bilateral riMLF and the PC were presumed to have been disturbed to the right, and this case contributed to the understanding of oculomotor disorders. We also investigated the characteristics of vertical eye movement disorders caused by cerebral hemorrhage.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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