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Case Report

Peduncular Hallucinosis 7 Months After Pontine Hemorrhage With Hypertrophic Olivary Degeneration: A Case Report

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HIGHLIGHTS

- Peduncular hallucinosis (PH) is a rare type of hallucination with colorful images.
- We report a case of PH after spontaneous pontine hemorrhage.
- We suggest correlation between hypertrophic olivary degeneration and PH.

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Peduncular Hallucinosis 7 Months After Pontine Hemorrhage With Hypertrophic Olivary Degeneration: A Case Report

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ABSTRACT

Peduncular hallucinosis is a rare type of hallucination, wherein patients see colorful and vivid images. It usually appears after damage to the midbrain, pons, or thalamus. We report the case of a 56-year-old man with peduncular hallucinosis after conservative care for spontaneous pontine hemorrhage, 7 months prior to presentation. He was treated with atypical antipsychotics, which resolved the symptoms. We suggest that it is important to consider peduncular hallucinosis in patients after injuries in subcortical areas and the brainstem. Additionally, we found changes in the hypertrophic olivary degeneration using magnetic resonance imaging, and we suggest the possibility of their correlation with peduncular hallucinosis.

Keywords: Hallucinations; Pons; Olivary Nucleus; Hemorrhagic Stroke

INTRODUCTION

Peduncular hallucinosis (PH) was first reported as a hallucination by Lhermitte in 1922 in patients with damage to the rostral brainstem or thalamus [1]. Patients with PH report seeing vividly colored animals and people that are smaller than they are. They also report rarely feeling threatened because they are aware that the images are not real [2]. To date, the mechanisms involved and treatment methods for PH have not yet been elucidated.

PH in patients with pontine hemorrhage has been reported in only a few cases. Herein, we report a case of PH that occurred 7 months after spontaneous pons hemorrhage and received conservative treatment.

Funding

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Conflict of Interest

Two co-authors of this manuscript is an editor of *Brain & NeuroRehabilitation*. The author did not engage in any part of the review and decision-making process for this manuscript. The other authors have no potential conflicts of interest to disclose.

Informed consent

We have obtained informed consent from the patient.

CASE REPORT

A 56-year-old man had been diagnosed with a spontaneous right pontine hemorrhage on a computed tomography image (**Fig. 1**), 7 months ago, at our hospital. He had sequelae such as left facial palsy, left hemiplegia of the upper and lower extremities, swallowing disorders, dysarthria, diplopia, balance disorders, tinnitus, and hearing loss. He had a history of hypertension, diabetes mellitus, and benign prostate hyperplasia. The patient was discharged from our hospital after receiving acute-phase treatment.

After discharge from our hospital, he received inpatient treatment at a local rehabilitation hospital before being referred back to our hospital due to a psychological problem for 2 weeks. He claimed that he saw hallucinations of people coming to his bed every night. At night, before sleeping, he saw a hooded grim reaper walking around, and during the day, with daytime somnolence, he saw corpses slightly smaller than ordinary people and people with flashy makeup driving around in wheelchairs. He also described the hallucination of a puppy that sparkled and changed color, which disappeared when he turned his head to observe it closely. However, since there were no responses when he spoke to the corpses, he soon realized that they were hallucinations. At the emergency room, neurological examination revealed left hemiplegia, grade 4 left upper limb muscle strength, grade 3 left lower limb muscle strength, and no sensory disturbance. A cranial nerve examination revealed right one-and-a-half syndrome, pendular nystagmus, left central facial palsy, tinnitus in the right ear, and left hearing loss. Right-sided ataxia was confirmed by cerebellar function tests. From these results, we concluded that the previous pontine hemorrhage was responsible for the symptoms.

Magnetic resonance imaging (MRI) confirmed changes suggestive of hypertrophic olivary degeneration (HOD) of the right ventral cord compared to the previous study (**Fig. 2**). However, there were no other lesions that support other psychological symptoms present on the MRI (**Fig. 3**). There were no symptoms associated with HOD, such as palatal tremor.

Serological examination revealed no signs of infection or electrolyte disorder. Electroencephalogram confirmed no epileptiform discharges, which ruled out temporal lobe epilepsy with hallucinations. Decreases in disorientation over time resulted in a score of 28 points on the Mini-Mental State Examination and 0 points on the Clinical Dementia Rating Scale, which suggested no significant cognitive decline or disorientation.

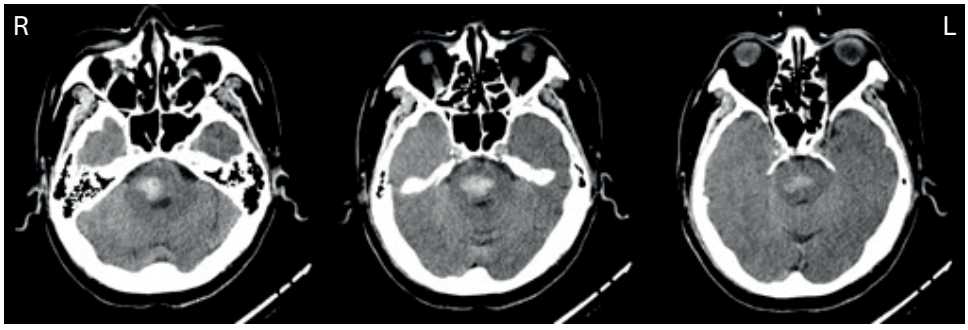

Fig. 1. Initial CT image of spontaneous right pontine hemorrhage.
CT, computed tomography.

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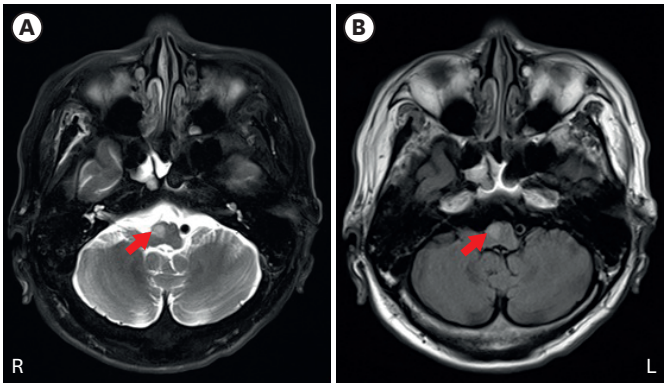

Fig. 2. Red arrow indicates increased signal intensity in the right olivary nucleus which indicates hypertrophic olivary generation due to pontine hemorrhage in (A) Axial T2-weighted image, (B) Axial T2-FLAIR image.

The hallucinations were vivid, and colorful images covered the entire visual field. It was a purely visual hallucination, and the patient was aware that the vision did not exist. Parkinson's disease, narcolepsy-cataplexy syndrome, delirium tremens, Lewy body dementia, and temporal lobe epilepsy may present such hallucinations. However, all tests, except imaging of the cranial nerves, were normal. No underlying disorders or adverse effects of medication were identified. Since all other possible causes, such as underlying diseases, medication, or trauma, were excluded, the diagnosis of PH due to a previous pontine hemorrhage could be made.

We prescribed haloperidol 1.5 mg twice daily to resolve the hallucination for 5 days. On the fifth day of admission, we concluded that haloperidol was ineffective and switched haloperidol to risperidone 0.125 mg once daily. After adjusting the medications, the hallucinations gradually improved and disappeared by the tenth day. After discharge, no further hallucinations were reported during the 10 months of outpatient follow-up.

DISCUSSION

Pontine hemorrhage accounts for 5%–20% of spontaneous cerebral hemorrhages and may be accompanied by coma, quadriplegia, loss of light reflex, aggressive sinusitis, respiratory arrest, and sideways gaze [3]. PH is rarely reported in patients with pontine hemorrhage [2,4]. This can be misdiagnosed because the mechanisms, diagnosis criteria, and treatment methods of PH in pontine hemorrhage are unclear. The lesions associated with PH may variably include the following structures at the midbrain level: the reticular formation; medial lemniscus and spinothalamic tract; raphe nuclei and periaqueductal grey; and finally, the tegmentum, cerebral peduncles, and pons [5-9]. At the diencephalic level, the paramedian thalamic region and the pulvinar are often included [8].

Although the mechanism of PH has not yet been elucidated, its pathophysiology is believed to be 2-fold. First, there are abnormalities in the ascending reticular activation system (ARAS), which is responsible for consciousness and rapid eye movement (REM) sleep [10]. Due to abnormalities in serotonin and cholinergic neurotransmitters in the dorsal raphe nucleus located in the bridge, the ponto-geniculo-occipital wave increases, REM sleep is overly induced, and ARAS is disrupted [11]. Midbrain and pons lesions involving the dorsal raphe nucleus result in loss of ascending serotonergic inhibition to the dorsal lateral geniculate

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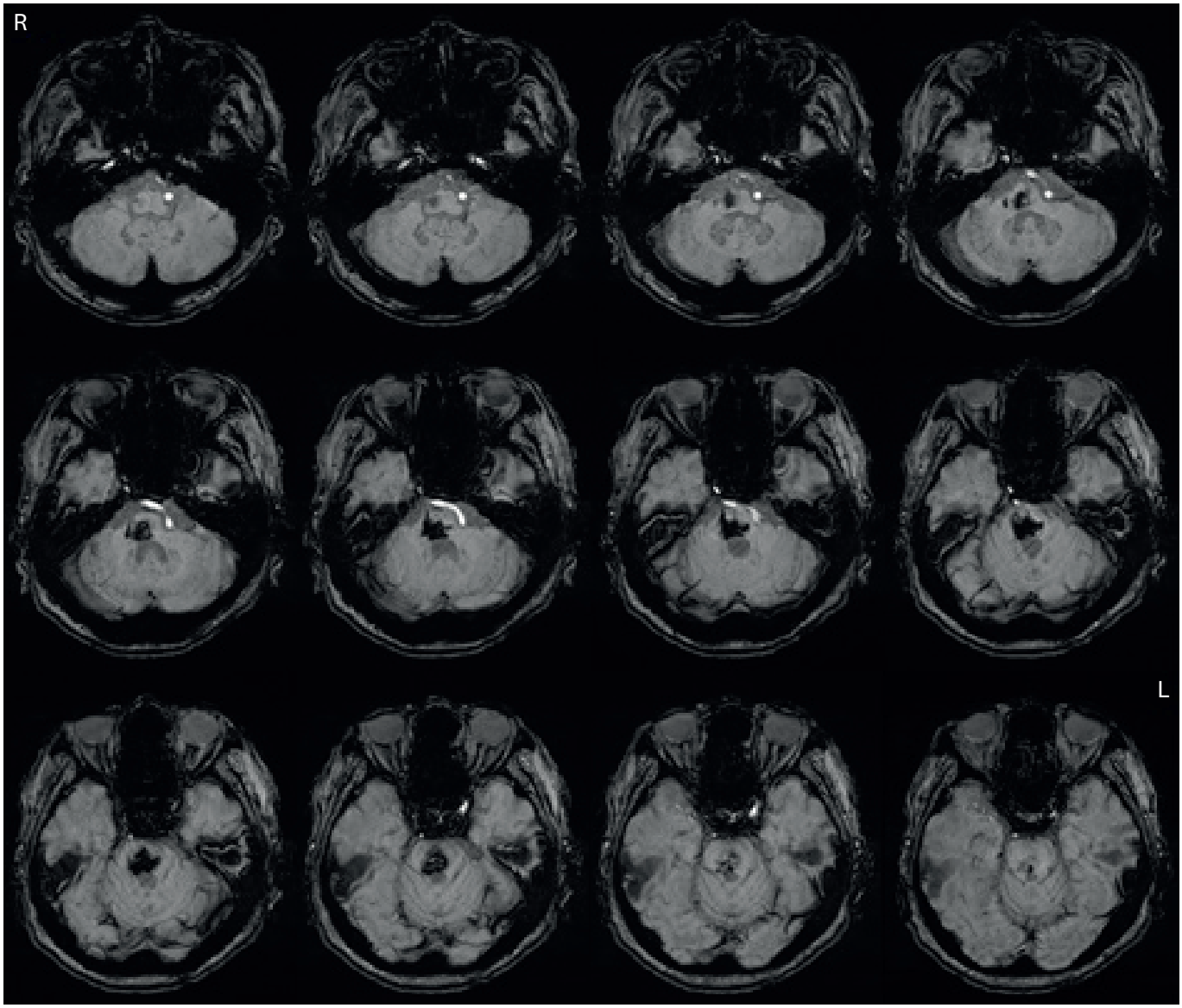

Fig. 3. No interval change of focal hypointense signals of the right pons with atrophic change and signal loss on SWI, suggesting resolving intracranial hemorrhage, leukomalacia change and hypertrophic olivary degeneration due to pontine hemorrhage.
SWI, susceptibility-weighted imaging.

nucleus. Consequently, a hyper-excited geniculate can generate visual hallucinations at the cortical level [12,13]. Second, due to damage to the basal ganglia, there is an abnormal response in the Meyer loop, which is responsible for visual signals. When an abnormality occurs in the Meyer loop, which connects the basal ganglia and inferior temporal lobe, substantia nigra compression or globus pallidus infarction can distort the visual signal [14].

Treatment methods for PH have not been established, but atypical antipsychotics, selective serotonin reuptake inhibitors, and anticonvulsants have been used in previously reported cases. Olanzapine and quetiapine have been reported to improve outcomes [15], and carbamazepine provides rapid symptom relief [16]. In some cases reported in Korea, improvement was reported after the administration of citalopram, olanzapine, or risperidone [4]. However, some studies have confirmed the complete disappearance of symptoms without medication [2].

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Compared to previous studies, our case report reflects the following common features of PH. The content of the hallucination was purely visual, with vivid and colorful visions that appeared in the entire field of vision, and the patient realized that these visions did not exist. It is known that PH can be present in Parkinson's disease, narcolepsy-cataplexy syndrome, delirium tremens, Lewy body dementia, and temporal lobe epilepsy, but he scored 28 on the Mini-Mental State Examination, and other tests except MRI were normal, which confirmed no association with the underlying disease. Since there was no other underlying disease or medication or traumatic history that could be the reason for the PH, we could conclude the previous hemorrhage was the cause of PH. However, compared to previous studies, HOD was found in this case. HOD is an enlargement of the inferior olivary nucleus and is rarely found following damage to the Guillain-Mollaret triangle (GMT) area [17,18]. It has been reported in some cases after pontine hemorrhage [19,20]. Pathologically, the most significant hypertrophy occurs 8.5 months after onset [21], and radiographically, it is observed on MR images 6 months after onset and disappears after 3 to 4 years [18]. Clinically, HOD has been reported to be indicative of palatal tremors and ocular myoclonus [22]. HOD is known to result from the ipsilateral central tegmental tract or contralateral olivary nucleus [19].

The pedunculopontine nucleus (PPN), thought to be associated with the development of PH, is located in the superior pons in the dorsolateral portion of the ponto-mesencephalic tegmentum [23]. It is responsible for the sleep cycle [24] and has been reported to cause vivid dreams and hallucinations when injured [25]. In addition, animal studies have shown that the dental nucleus (DN) is the nucleus most activated during PPN deep brain stimulation [26]. The DN is a component of the GMT that forms the dentatorubral olivary pathway, which is involved in the mechanism of HOD generation.

To date, no clinical association between PH and HOD has been reported. However, both PH and HOD are rarely reported as clinical findings, and it is difficult to confirm the absence of a relationship between the 2 as clear mechanisms or symptoms have not yet been elucidated. In other words, there is a possibility that the functional deterioration of the PPN causes HOD by affecting the DN, or conversely, PH can occur due to the occurrence of HOD, which causes functional deterioration of the PPN through the GMT. In addition, the period from brainstem damage to the onset of PH has been observed to vary from several months [8] to 2.5 years [4] after onset, which is similar to the period in which HOD appears. We cannot confirm whether the pontine hemorrhage-damaged PPN and central tegmental tract independently resulted in PH and HOD, or if the HOD affected PPN and resulted in PH, or if damage to the PPN affected RN and resulted in HOD. Follow-up studies and more case reports on the PH-HOD relationship are needed.

PH can be suspected in patients with brainstem damage who experience vivid hallucinations of colorful animals or people, especially when the hallucinations can be recognized as such by the patients themselves. Patients with pontine hemorrhage require special attention, as they are often accompanied by dizziness, eye movement disorders, and dysarthria. Their symptoms, such as hallucination, can be easily taken lightly. Since the exact pathophysiology of this phenomenon is still unknown, there is no exact criteria for diagnosis or treatment. We suggest one possible mechanism of PH by HOD, but further studies, including more case reports, are needed to understand the phenomenon.

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