



Case Report

Peduncular hallucinosis: A polysomnographic and spect study of a patient and efficacy of serotonergic therapy

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ABSTRACT

Peduncular hallucinosis (PH) consists of formed and coloured visual images, which the patient knows are unreal; it is often associated with lesions of the pons, midbrain and diencephalon. A 72-year-old man had noted the sudden onset of visual hallucinations one year before, specifying the time and body position in a 4-week, 24-h diary. Thereafter, he underwent video-polysomnography (VPSG), brain magnetic resonance imaging (MRI), angiography (MRA), proton spectroscopy (¹H MRS), and single photon emission tomography (SPECT). Patient's diaries and VPSG showed a strong clustering of hallucinatory experiences during the evening/night time while lying in supine position, similar to hypnagogic hallucination and sleep paralysis in supine position. Repeated episodes of REM sleep behaviour disorder (RBD) occurred during the night. MRI and MRA showed an elongated and dilated left internal carotid artery displacing the left subthalamus upwards, and ¹H MRS relatively decreased *N*-acetyl-aspartate in the left subthalamus. Brain SPECT during PH revealed hypoperfusion in the right temporal region and hyperperfusion in the left occipital and right opercular regions (the latter possibly related to the patient's awareness of unreality). PH resolved with serotonergic (citalopram) therapy.

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1. Introduction

Peduncular hallucinosis (PH) is characterized by visual hallucinations (which the patient is aware are unreal) usually consisting of moving animals or people and more pronounced at dusk [1].

We examined a patient with long lasting, vividly formed hallucinations recurring especially in supine posture and in the evening.

2. Case report

A 72-year-old man had noted the sudden onset of visual hallucinations one year before examination. He described the hallucinations as complex visual scenes, elaborated and rich in colours, extremely realistic and consisting of landscapes, animals and unfamiliar persons, occupying any part of the visual field, recurring quite regularly in the evening, especially when he was lying on his back resting. The patient denied hallucinations when he was in a prone and/or lateral position. From the same time, dreams were also reported as more vivid and emotionally charged. The pa-

tient was neurologically normal. Daytime EEG and neuropsychological examination (Beck Depression Inventory, MMSE, Frontal Assessment Battery) were also normal.

CSF tau, 14-3-3 and hypocretin levels, and dopamine transporter brain single photon emission computerised tomography (SPECT) imaging were normal. HLA typing for DQB1*0602 was negative.

3. Methods

For 4 weeks the patient was asked to fill in a 24-h diary detailing the time of day and the body position associated with the hallucinations. VPSG, MRI, MRA, ¹H MRS with a point-resolved proton spectroscopy sequence (PRESS, TR = 2000 ms, TE = 272 ms) with a 1.5 × 1.5 × 1.5 cm volume of interest placed in the diencephalon (see Section 4) of each cerebral hemisphere were performed.

Brain SPECT was obtained by intravenous injection of 740 MBq ethyl-cysteinate-dimer labelled with technetium-99m (99mTc) at 6:00 P.M. when the patient was in bed, in supine position, in a dimly lit room, and experiencing visual hallucinations, after 50 min (when visual hallucinations were still occurring). Image acquisition started 10 min later.

Control SPECT was obtained one week later but in the absence of hallucinations.

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Image registration and subtractions of ictal (with hallucinations) and interictal (without hallucinations) SPECT were performed with the previously described ISAS (Ictal-Interictal SPECT Analysis by SPM, <http://spect.yale.edu>) method.

4. Results

4.1. Four-week 24-h diary analysis

Analysis of the patient's diaries showed that hallucinations occurred in the evening/night time, between 8:00 P.M. and 5:00 A.M., when the patient was lying on his back. Supine posture during morning/afternoon time was not, however, associated with hallucinations (Fig. 1A). Moreover, the prone and/or lateral decubitus positions were reported as not associated with the occurrence of hallucinations.

4.2. VPSG

During relaxed wakefulness, and while the patient was experiencing hallucinations, EEG showed a posterior alpha rhythm at 8–9 c/s and some theta activity over the central regions associated with irregular conjugate eye movements, eyelid fluttering, and spontaneous sympathetic skin response (SSRs) (Fig. 1B). Hallucinations continued for nearly two hours before sleep onset (Fig. 1B). During REM sleep, three episodes occurred which were consistent with RBD; SSRs were now absent. Spontaneously awakened after the third RBD episode, the patient reported a dream in which he was dealing with a prostitute (Fig. 1B).

4.3. MRI, MRA and ¹H MRS

MRI showed small hyperintense focal lesions of ischemic nature in the left subinsular region and MRA upward displacement of the left subthalamic region by an elongated and dilated left internal carotid artery and its bifurcation (Fig. 1C). Diffusion weighted abnormalities were absent. ¹H MRS showed relatively decreased NAA and Cho peaks in the left subthalamus (NAA/Cr 0.65; Cho/Cr 0.33) as compared with the right subthalamus (NAA/Cr 1.28; Cho/Cr 0.75).

4.4. Brain SPECT

On brain SPECT, during visual hallucinations two clusters of significant hyperperfusion were observed in the left middle occipital gyrus (MNI coordinates $x = -40$, $y = -84$, $z = 12$) and the right inferior frontal-rolandic opercular gyri (MNI coordinates $x = 52$, $y = 6$, $z = 10$). An area of hypoperfusion was present in the right middle temporal gyrus (MNI coordinates $x = 52$, $y = -40$, $z = -12$; Fig. 1C).

4.5. Treatment

Clonazepam treatment up to 2 mg/day gave only partial relief. Citalopram 20 mg/day was then added and after twenty days the hallucinations were completely abolished. Hallucinations were still absent after one year while on citalopram plus clonazepam treatment. Repeated neurological and neuropsychological assessments were normal.

5. Discussion

Our patient had a sudden onset of visual hallucinations recurring late in the day, particularly at dusk when he was lying on

his back, and persisting for over one year in the absence of any other pathology. The visual hallucinations fit the criteria of true hallucinatory experiences [2] and were consistent with a diagnosis of PH [2] recurring with a circadian rhythm at evening–night time as a hypnagogic experience. Remarkably VPSG disclosed the presence of several episodes of RBD, a finding not previously reported in PH.

MRI and MRA showed an elongated and dilated left internal carotid artery displacing the left subthalamus upwards, and ¹H MRS showed a relative decrease of NAA and Cho peaks in the left subthalamus. It is tempting to attribute our patient's symptoms to a vascular compression of the subthalamus, especially since subthalamic lesions, already reported in PH [3], could dysregulate several interconnected loops, thus releasing dissociated motor (RBD) and hallucinatory phenomena.

SPECT in our patient showed hyperperfusion in the left occipital and right opercular regions and hypoperfusion in the right temporal region. Unilateral activation of the left middle occipital gyrus was unexpected, but asymmetric activation with left-hemisphere predominance has been reported in studies of mental imagery [4] or related to the content of hallucinations [5].

The right rolandic operculum activation may be related to the detection and attempt to resolve various conflicting signals [6] or to the patient building proper judgement about his complex hallucinations [7]. The right temporal lobe hypoperfusion is similar to the findings in medication-induced hallucinations in parkinsonian patients [8].

Finally, PH resolved with serotonergic therapy (citalopram plus clonazepam), which is in line with the concept that serotonergic impairment is an important modulating factor of hallucinations [2,9].

We wish to emphasize how PH in our patient was linked to the hypnagogic period and reminiscent of the hypnagogic phenomena. As in sleep paralysis, which is often accompanied by hypnagogic hallucinations [10], body position and timing appeared to be two constant situational factors affecting PH in our patient. Dim ambient lighting and the reduced vigilance levels were probably other contributing factors. PH could therefore represent an instance of a physiological phenomenon (hypnagogic hallucinations) amplified in its recurrence and intensity by specific CNS lesions.

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