https://doi.org/10.22365/jpsych.2023.026 / Psychiatriki 2024, 35:78–82

Case report

Peduncular hallucinosis associated with pontine hemorrhage in an adult patient

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ARTICLE HISTORY: Received 30 May 2023/Revised 11 August 2023/Published Online 14 November 2023

ABSTRACT

Peduncular hallucinosis refers to a rare neurophychiatric disorder presenting with vivid visual hallucinations, disturbances of sleep, and oculomotor dysfunction. It is typically caused by mesencephalic lesions. Nonetheless, a few cases have also been reported, in which the same syndrome was associated with thalamic and pontine lesions. We report the case of a 63-year-old male patient presenting to the Emergency Department of our hospital with irritability, gait difficulty, and diplopia of sudden onset two hours ago. Neurological examination revealed dysarthria, right facial palsy, bilateral gaze palsy, dysmetria of his left extremities, left-sided hemihypaethesia and extensory plantar response on the left. Brain computerized tomography (CT) showed a hemorrhagic lesion on the right lateral side of the pons. During his hospitalization at the Department of Neurology, he developed visual hallucinations, confusion, disorientation, insomnia, and strong emotional response. An extensive laboratory screening was performed and showed no abnormal findings. Suspecting peduncular hallucinosis due to the brainstem lesion, treatment with quetiapine and melatonin was administered to the patient and symptoms resolved completely within days. Subsequently, gradual neurological clinical improvement was also noted and two weeks after his admission, a repeated brain CT and a brain magnetic resonance imaging (MRI) showed partial absorption of the brainstem hemorrhage. The patient underwent rehabilitation for two months, showing further clinical improvement, and treatment with quetiapine and melatonin was discontinued without any further episodes being noted. A repeated brain MRI was performed two months after his admission to our hospital and showed no hemorrhage, but a mixed signal intensity core and a hypointense hemosiderin rim at the location of the absorbed hemorrhagic lesion, compatible with pontine carvenoma. Peduncular hallucinosis is most commonly associated with ischemic lesions of the posterior brain blood circulation, but different lesions have been reported, like vasospasm, brain tumors, encephalitis, hemorrhage associated with vascular malformations, such as a carvenoma, as seen in our case, representing a very rare form of peduncular hallucinosis.

KEYWORDS: Peduncular hallucinosis, hemorrhage, pons, carvenoma, case report.

Introduction

Peduncular hallucinosis refers to a rare neurophychiatric disorder presenting with vivid and dream-like visual hallucinations, disturbances of sleep, and oculomotor dysfunction.[1] The disorder is usually associated with lesions of the midbrain. However, thalamic and pontine lesions have also been related, less frequently, to visual hallucinations and brainstem dysfunction. Moreover, ischemic lesions of the posterior brain blood circulation have been described as the most common cause of peduncular hallucinosis, but different underlying etiologies have also been reported, e.g., vasospasm, brain tumors, encephalitis, subarachnoid hemorrhage, and vascular malformations. Confirming the diagnosis requires the exclusion of other possible causes of delirium in these patients and, once diagnosed, treatment with atypical antipsychotic medications is recommended.[2,3]

The aim of our paper is to report the challenges of diagnosis and treatment that a very rare case of hallucino-

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sis posed in an adult patient after pontine hemorrhage due to a carvenous malformation.

Case Presentation

A 63-year-old man presented to the Emergency Department of our hospital with sudden onset of irritability, gait difficulty, and diplopia of sudden onset two hours ago. Apart from smoking (18 pack years), he reported a free medical history and he was not on medications. Personal or family history of mental illness, alcohol or psychotropic substances abuse was denied. The patient's vital signs were normal. Neurological examination, however, revealed dysarthria, right facial palsy, bilateral horizontal gaze palsy, muscle weakness and dysmetria of his left extremities, left-sided hemihypaethesia and extensory plantar response on the left. Routine blood and urine tests, as well as a chest x-ray, were unremarkable. A computerized tomography (CT) and angiography (CTA) of his brain were performed, which showed a hemorrhagic lesion located dorsally at the tegmental area of the pons (figure 1a), without findings of vascular malformations.

During his hospitalization at the Department of Neurology, within five days of his admission, he developed symptoms resembling acute delirium. He had visual hallucinations of rats and spiders climbing down the wall. Twice, he was also seen interacting with deceased family members, having conversations and handshaking with them. While the latter episodes imply the presence of multimodal hallucinations with an additional auditory and/or tactile component, such details regarding the exact nature of his hallucinations could not be provided by the patient for the particular episodes. During these episodes, the patient was also confused, disorientated and displaying exaggerated emotional responses. In contrast, in between these episodes, the patient was oriented, demonstrating a linear and organized thought process as well as appropriate affect. In addition, he was able to describe the episodes. The patient's hallucinations occurred twice daily, mostly at night. He also complained about insomnia

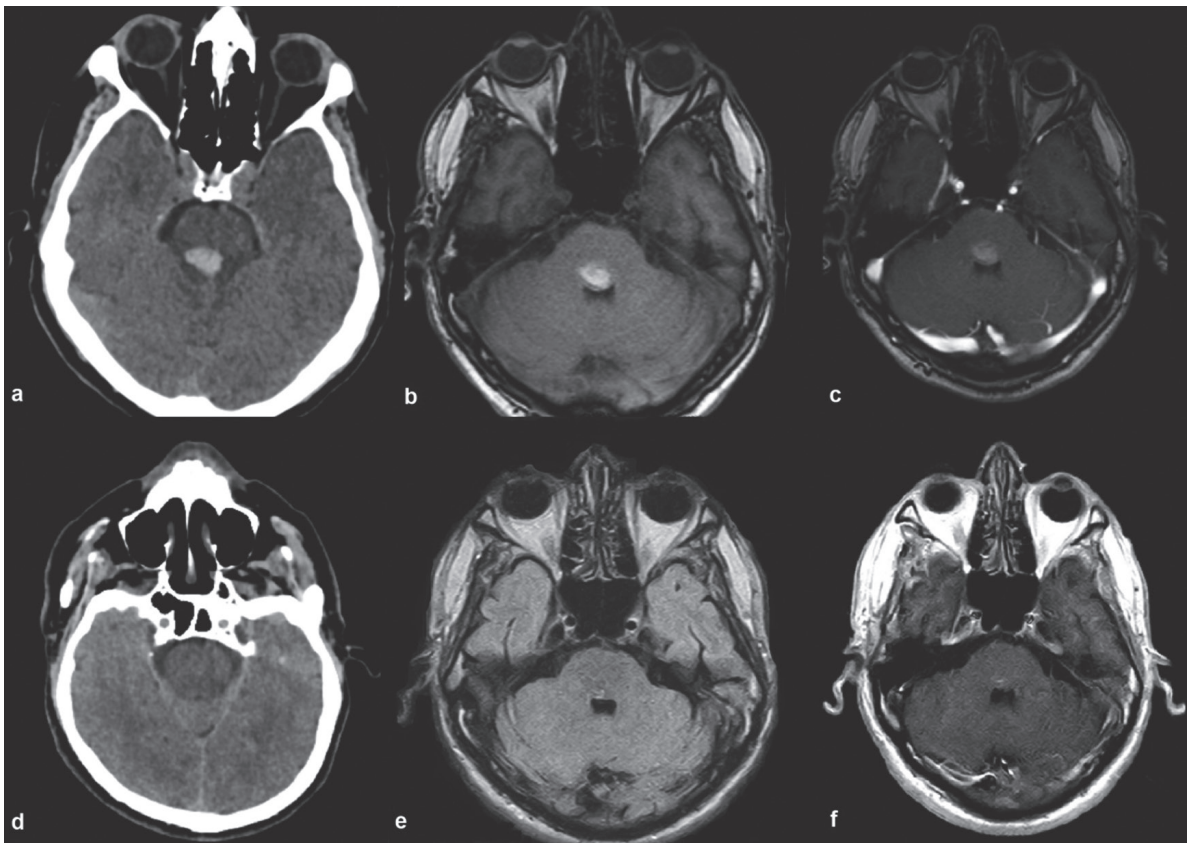

Figure 1. (a) Non contrast axial brain computerized tomography showing a hemorrhagic lesion at the tegmental area of the pons; (b) Axial brain magnetic resonance imaging T2 fluid-attenuated inversion recovery (FLAIR) sequence; and (c) T1 sequence with gadolinium, two weeks after pontine hemorrhage, showing partial absorption of the hemorrhagic lesion, (d) Non contrast axial brain computerized tomography; e. Axial brain magnetic resonance imaging T2 fluid-attenuated inversion recovery (FLAIR) sequence, and f. T1 sequence with gadolinium, two months after pontine hemorrhage, showing absorption of the hemorrhage.

and irritability before night sleep. Oral drops of haloperidol were administered to the patient, to no avail. Electroencephalography and electrocardiography were performed at that time and showed no abnormal findings. An extensive laboratory screening was performed to exclude other possible causes, including a lumbar puncture (2 white cells/cm^3, 0 red blood cells, normal protein and glucose levels, negative culture), as well as blood and urine tests (complete blood count, urine toxicology screen, blood sugar, urea, creatinine, hepatic function, ferritin, vitamin B12, thiamine, folic acid, thyroid-stimulating hormone, free T4, T3, protein and immune electrophoresis, anti-nuclear antibodies, erythrocyte sedimentation rate, C-reactive protein, blood and urine cultures, serology for Treponema pallidum. hepatitis B virus, hepatitis C virus and human immunodeficiency virus), lacking abnormal findings. Taking into account his recent brainstem lesion, peduncular hallucinosis was considered a probable cause of the delirium and the patient was treated with 25 mg of quetiapine twice daily and 4mg of melatonin once daily, orally, at bedtime. Within three days, symptoms resolved completely and the patient returned to his baseline mental state.

The patient gradually improved regarding his dysarthria, muscle weakness, dysmetria (most likely attributed to muscle weakness due to corticospinal tract lesion) and oculomotor dysfunction. Two weeks after his admission, a repeated brain CT and a brain magnetic resonance imaging (MRI) were performed and showed partial absorption of the brainstem hemorrhage (figure 1b and c). The patient was referred to the Department of Rehabilitation of our hospital and after two months (two days before his discharge from our hospital), he was re-evaluated at our department. Neurological examination revealed only mild lateral gaze palsy and a repeated brain CT and MRI, before his discharge (figure 1d, e and f), showed no hemorrhagic lesion but a mixed signal intensity core and a hypointense hemosiderin rim at the location of prior hemorrhage, compatible with pontine carvenoma (figure 2). Thus, neurosurgical consultation was sought; however, resection of the carvenoma was not suggested. Treatment with quetiapine and melatonin was gradually discontinued within the next month, yet recurrence of hallucinosis was not reported.

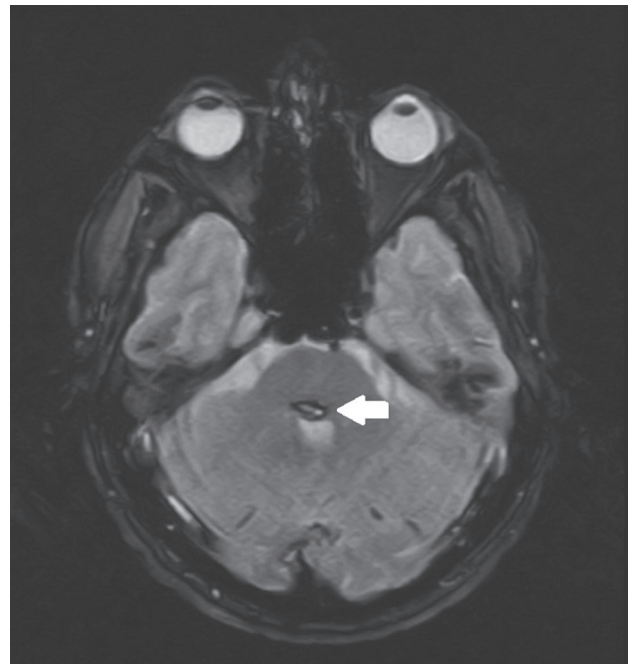

Figure 2. Axial brain magnetic resonance imaging two months after pontine hemorrhage: Susceptibility weighted imaging (SWI) sequence showing a pontine mixed signal intensity core and a hypointense hemosiderin rim (arrow).

Discussion

Peduncular hallucinosis is typically described as complex visual hallucinations with realistic, dynamic scenes, often involving familiar people or places. Patients have difficulty distinguishing their hallucinations from reality during the episodes, although it is not unusual to have insight into the hallucinations, as seen in our case. According to clinical descriptions, these hallucinations are so vivid that most patients will start interacting, either verbally or physically, with people or other parts of the environment perceived during their hallucinations.[3] Hallucinations can occur at any time of the day but are more frequent at night. Between hallucinations, patients have intact memory and are able to describe their hallucinations accurately.[4] Except for vivid and dream-like hallucinations, other frequent associated clinical findings include symptoms and signs of brainstem dysfunction, such as ocular motor impairment, cerebellar dysfunction, as well as sleep and arousal disturbances.[3,5]

The exact pathophysiology of peduncular hallucinosis is unknown. Various possible mechanisms have been proposed, most of them involving dysfunction of the brainstem reticular formation, deregulation of the thalamic of sensory input thresholds due to disruption of excitatory cholinergic (arising from the pontine tegmentum) and inhibitory serotonergic (arising from the dorsal raphe nuclei) brainstem neurotransmission, hence promoting overexcitation of the dorsal lateral geniculate nucleus of the thalamus, which is involved in higher order visual processing, thus resulting in visual hallucinations, and in rare instances, additionally in auditory and/or tactile hallucinations. Furthermore, it is known that the dorsal raphe nucleus is also re-

sponsible for the total sleep cycle (REM and non-REM sleep), which could explain sleep disturbances in these patients (nighttime wakefulness and hypersomnolence during the day) and correlate with the other clinical findings of the disease.[5–7]

Peduncular hallucinosis associated with vascular malformations has been previously described in the literature. We found four case reports, including six patients with peduncular hallucinosis due to aneurysms in the posterior brain blood circulation,[8–11] and only one case due to a pontine carvenoma.[6] In all those cases, hemorrhage was present due to rupture of the vascular malformations, as seen in our patient, had occurred. In contrast to our case, pontine carvenoma had already been previously diagnosed in the case described by Couse et al[6] and symptoms of peduncular hallucinosis developed after brainstem hemorrhage.

Conclusion

To our knowledge, this is the second case report of peduncular hallucinosis associated with pontine hemorrhage due to a brainstem carvenoma. We suggest that clinicians encountering patients with hallucinations, sleep disorders and neurological dysfunction, should consider peduncular hallucinosis as a diagnosis when other possible causes have been excluded.

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Παρουσίαση περίπτωσης

Σκελική ψευδαισθήτωση σχετιζόμενη με αιμορραγία γέφυρας σε ενήλικο ασθενή

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ΙΣΤΟΡΙΚΟ ΑΡΘΡΟΥ: Παραλήφθηκε 30 Μαΐου 2023/Αναθεωρήθηκε 11 Αυγούστου 2023/Δημοσιεύθηκε Διαδικτυακά 14 Νοεμβρίου 2023

ΠΕΡΙΛΗΨΗ

Η σκελική ψευδαισθήτωση αφορά σε μια σπάνια νευρολογική διαταραχή, η οποία χαρακτηρίζεται από ζωηρές οπτικές ψευδαισθήσεις, που σχετίζονται με διαταραχές του ύπνου και της οφθαλμοκινητικότητας. Σπάνια οφείλεται σε βλάβες εκτός των εγκεφαλικών σκελών, όπως ο θάλαμος και η γέφυρα. Παρουσιάζουμε την περίπτωση ενός ασθενούς 63 ετών, με ιστορικό καπνίσματος, ο οποίος νοσηλεύτηκε στο Νευρολογικό Τμήμα του νοσοκομείου μας, λόγω αιφνίδιας εγκατάστασης διαταραχής όρασης, δυσχέρειας βάδισης και ψυχοκινητικής ανησυχίας, στα πλαίσια ενδοεγκεφαλικής αιμορραγίας στο δεξιό πλάγιο τμήμα της γέφυρας, η οποία αναδείχθηκε σε επείγουσα αξονική τομογραφία. Η νευρολογική του εξέταση ανέδειξε δυσαρθρία, αμφοτερόπλευρη πάρεση πλάγιων συζυγών οφθαλμικών κινήσεων, πτώση γωνίας στόματος δεξιά, υπαισθησία αριστερού ημισώματος, δυσμετρία αριστερών άκρων, καθώς και σημείο Babinski αριστερά. Εντός της νοσηλείας του, παρουσίασε οπτικές ψευδαισθήσεις, σύγχυση, αποπροσανατολισμό, διαταραχή συναισθήματος και συμπεριφοράς, καθώς και διαταραχές ύπνου. Διενεργήθηκε εκτεταμένος εργαστηριακός έλεγχος, χωρίς την ανάδειξη άλλου πιθανού αιτίου οργανικού ψυχοσυνδρόμου και ο ασθενής αντιμετωπίστηκε επιτυχώς με θεραπευτική αγωγή κουετιαπίνης και μελατονίνης, εντός ημερών. Προοδευτικά, παρατηρήθηκε κλινική βελτίωση της νευρολογικής εικόνας, ενώ από τον επαναληπτικό έλεγχο με μαγνητική τομογραφία εγκεφάλου διαπιστώθηκε μερική απορρόφηση της αιμορραγίας. Ο ασθενής συνέχισε την αποκατάστασή του για δύο μήνες και παρουσίασε περαιτέρω κλινική βελτίωση, ενώ προοδευτικά διεκόπη η θεραπευτική αγωγή, χωρίς υποτροπή των συμπτωμάτων. Έπειτα από δύο μήνες, έγινε επανεκτίμηση του ασθενούς και διενεργήθηκε εκ νέου απεικονιστικός έλεγχος με μαγνητική τομογραφία εγκεφάλου, όπου διαπιστώθηκε απορρόφηση της αιμορραγίας και παρουσία δακτυλίου αιμοσιδηρίνης εντός της τοποθεσίας της αιμορραγίας, στα πλαίσια σηραγγώδους αιμαγγειώματος στη γέφυρα. Η ισχαιμία της οπίσθιας κυκλοφορίας αποτελεί την πιο συχνή αιτία σκελικής ψευδαισθήτωσης, αν και έχουν αναφερθεί σπανιότερα αίτια, όπως μάζες εγκεφάλου, αγγειόσπασμος, εγκεφαλίτιδα και αιμορραγία σχετιζόμενη με αγγειακές δυσπλασίες, όπως το σηραγγώδες αγγείωμα, που παρατηρήθηκε στην περίπτωσή μας και αντιπροσωπεύει μία πολύ σπάνια μορφή σκελικής ψευδαισθήτωσης.

ΛΕΞΕΙΣ ΕΥΡΕΤΗΡΙΟΥ: Σκελική ψευδαισθήτωση, αιμορραγία, γέφυρα, σηραγγώδες αιμαγγείωμα, παρουσίαση περιστατικού.

Συγγραφέας επικοινωνίας: Μιχαήλ Παπαντωνίου, Νευρολογικό Τμήμα, Γενικό Νοσοκομείο Αθηνών «Γ. Γεννηματάς», Λεωφόρος Μεσογείων 154, 115 27 Αθήνα, Διεύθυνση e-mail: michael.papant@gmail.com