

Peduncular hallucinosis associated with a pontine cavernoma

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Abstract

Peduncular hallucinosis is a rare neurological disorder characterized by visual hallucinations, often described to be vivid and dream-like. While the exact pathophysiology has yet to be elucidated, most cases to date have suggested an etiology stemming from lesions to the thalamus or midbrain. Here presented is a case of a 54-year-old female with peduncular hallucinosis secondary to a pontine cavernoma hemorrhage in the setting of essential hypertension. The patient's vivid visual and auditory hallucinations aligned temporally with the lesion's discovery and resolved after pharmaceutical treatment. This case represents a rare form of peduncular hallucinosis secondary to a pontine cavernoma hemorrhage leading to vasospasm in the arteries feeding the brainstem.

Introduction

Peduncular hallucinosis (PH) is a rare neurological disorder characterized by vivid visual hallucinations (VH) which are often associated with disturbances of sleep and oculomotor function.^{1,2} PH is associated with a range of different pathologies of the central nervous system including: vascular and infectious lesions of the midbrain, pons and thalamus; local subarachnoid hemorrhage; compression by local and distal tumors; basilar migraines; basilar vascular hypoplasia and following regional surgical or angiographic interventions.³ Due its rarity and the array of conditions associated with its development, diagnosing PH presents a rare and unique challenge to the clinician. Here, we hope to add to the conditions associated with PH by presenting an exceptional case of PH resulting from a pontine hemangioma hemorrhage.

Case Report

A 54-year-old female with a past medical history of depression, anxiety, hypothyroidism and hypertension presented to an outside hospital after attempting suicide by cutting both of her wrists. While hospital-

ized, a medical work-up was performed due to the patient's recent weight loss. A CT brain identified a pontine cavernoma prompting the patient to be transferred to UC Irvine (UCI) for a neurosurgical evaluation nine days after her suicide attempt.

At UCI, the patient experienced both auditory (AH) and visual hallucinations (VH); most commonly the hallucinations presented as members of her family. She was observed numerous times to be conversing with imaginary family members and informed staff that she saw these family members in her room on several occasions. Additionally, the VH and AH were not always experienced together. For example, she experienced AH of a family member's voice emanating from another room resulting in confusion, disorientation and eliciting a strong emotional response. She was also seen to be interacting with her VH; for example, she was seen knitting with imaginary needles and petting an imaginary dog. The patient's hallucinations occurred once or twice daily and occurred mostly at night. In between these episodes, the patient was appropriately oriented, demonstrated a linear, organized thought process and had no abnormal thought content. The patient was able to describe many of her hallucinations in detail, often depicting them as being *surreal* and *dream-like*. During this time the patient also complained of insomnia, difficulty with ocular movements and vertigo.

On ophthalmic exam, an impairment was noted with her ocular motility. Using a 9 point ocular motility scale ranging from -4 to 4, she demonstrated a -2 restriction in all directions in her right eye and -1 restriction in all directions except -2 restriction of abduction in her left eye. No other abnormalities were noted on her ophthalmic, physical and neurological exam.

Work up conducted at UCI included a complete metabolic panel, complete blood count, urine toxicology screen, thyroid stimulating hormone level and a urine analysis, all of which were within normal limits. An MRI brain revealed a 1.2 cm region of hemorrhage within the posterior pons. Neurosurgery determined that the best course was to monitor the hemorrhagic lesion in lieu of evacuation. A diagnosis of peduncular hallucinosis was made and, in accordance with previous case reports, the patient was started on Olanzapine 5 mg at bedtime.³⁻⁵ The patient was also started on 20 mg fluoxetine daily for her depression, a scopolamine patch for her vertigo and continued on her home hydrochlorothiazide and levothyroxine. Thereafter the VH resolved within the next 24 hours and the patient was eventually discharged home in stable condition.

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Discussion

The patient described above reported new onset hallucinations that became noticeable to the medical staff shortly after she was stabilized following a suicide attempt. In keeping with the current literature, her hallucinations were vivid, complex and centered on persons familiar to her; specifically members of her family.² Other characteristics of her hallucinations were in accordance with previously published cases of PH, including: VH combined with AH,⁶ active interaction by the patient with her hallucinations,^{1,7} and the hallucinations predominately occurring at night.^{2,4,8} When not actively hallucinating, the patient demonstrated insight into the nature of her hallucinations, which correlates with some but not all reports of PH.^{2,8,9} Benke *et al.* proposed that the discrepancy in insight is due to the differences in the manner by which patients are interrogated about the episodes as well as the time lag between the hallucination and the evaluation by clinical staff. This may explain why our patient demonstrated insight only once her hallucinations had subsided. In addition to hallucinations, PH often presents with ocular motor and sleep disturbances;^{2,10} both of which our patient experienced while hospitalized. In a case series of five PH patients, Benke *et al.* reported that all five patients struggled with recalling information provided to them in a mental status exam. This finding was noted in our case as the patient scored a 24 on her

MOCA losing the majority of points in the section requiring her to recall words presented to her a few minutes earlier.

PH has been associated with a range of differing CNS pathologies;^{2,3,5,11,12} however, regardless of the etiology, lesions of midbrain and/or pontine structures appear to be crucial for the characteristic cognitive defects. The mechanism for PH implicates the brainstem reticular formation, its thalamic targets and the projections connecting the two. Briefly, excitatory, cholinergic projections arise from the pontine tegmentum and inhibitory, serotonergic projections arise from the dorsal raphe nuclei. Projections from both of these centers influence the dorsal lateral geniculate nucleus (LGN) of the thalamus. Brainstem lesions may disrupt the serotonergic, inhibitory raphe nucleus inputs resulting in excitation of the dorsal LGN and dysregulation of LGN projections to visual cortical regions leading to complex VHS. Similarly, lesions involving thalamic nuclei may disrupt the important processing function of these structures, resulting in impaired retinal signals and visual hallucinations. In addition to its involvement in inhibiting the LGN, the dorsal raphe nuclei have been implicated in both the sleep-wake cycle and regulation of REM and non-REM sleep.¹⁰ Due to this relation it has been proposed that the visual experiences in PH are theoretically similar to those of REM sleep.

Pontine cavernomas are a collection of malformed blood vessels and because of their fragility, hemorrhage is a common complication. Three previous reports have been published in which subarachnoid hemorrhage resulted in PH.^{5,13,14} In these cases, the proposed mechanism of PH onset begins with free blood irritating blood vessels leading to vasospasm resulting in reduced cerebral perfusion in the midbrain. The authors also proposed a less likely alternative, in which a more localized effect of pressure on the midbrain due to tension

in the interpeduncular cistern could also give rise to VH.^{5,13,14} In the case described by Harada, the authors argued that the two to three day delay in the onset of hallucinations was due to vasospasm rather than direct damage to the brainstem.

Conclusions

In the case here presented it is difficult to know if subtler symptoms began prior to the pontine cavernoma hemorrhage was discovered; however, what is clear is that there was a multiple day delay between the discovery of the hemorrhage and presentation of hallucinations. Taken together, the proposed pathophysiology of our case began with hemorrhage from her pontine hemangioma leading to vasospasm of the arteries feeding the brainstem resulting in disruption of the dorsal raphe nucleus and its inputs onto the dorsal LGN ending in visual hallucinations and PH.

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