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Case Report

Peduncular Hallucination Status Secondary to Thalamic Stroke: Brief Case Report and Review of the Literature

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Abstract

Background: The role of thalamus in the pathogenesis of visual and auditory hallucinations has been described as "Peduncular Hallucinosis". This kind of hallucinations has been reported to be associated with thalamic injury joined to midbrain structures damage. Hallucinations are filled with vivid images of animals and colorful characters in motion and in many cases are associated with sleep disorders. Despite these characteristics, patients generally distinguish them as not real.

Case Report: The present case has the distinction of having characteristics somewhat different from those described in published cases. In the first place, it is the case of the longest duration published up to date, 5 years, secondly, contrary to what usually happens, the patient does not criticize the objects and understands them as real

Conclusions: We present an updated review about literature published and redefinition of the concept the particular characteristics of this rare syndrome. When facing hallucinatory cases of an organic type, especially in patients with cardiovascular risk factors, it is important to rule out rare syndromes such as Charles Bonnet or hallucinations of thalamic origin.

Keywords: Perceptive Distortion; Peduncular Hallucinosis; Status; Thalamus; Thalamic Stroke; Visual Hallucinosis

Introduction

'Peduncular hallucinosis' (PH) is used to describe vivid and complex hallucinations in the presence of lesions of the midbrain and/or thalamus [1]. Since the first publication by Jean Lhermitte in 1922 of his paper on hallucinosis, peduncular type has been described as a purely visual phenomenon [2]. In this paper he described this rare syndrome as the cerebral peduncle cap syndrome describing psycho-sensory disturbances in midbrain lesions.

It was first described in a 72-year-old woman with hallucinations of strangely, colorfully attired people and groups of children which occurred at dusk. These were associated with no alteration of conscious state and neurological signs consistent with an infarct of the pons and midbrain [2]. Lhermitte's colleague Von Bogaert

coined the term 'peduncular hallucinosis' in his description of a patient with a similar presentation, together with post-mortem neuropathological evidence of midbrain infarction [3]. The term 'peduncular' was not intended as a reference solely to the cerebral peduncles but to the whole midbrain and its surrounds. De Morsier extended the syndrome to include thalamic lesions, and re-established the lesion model for PH [4] whereas his predecessors had suggested sleep-wake cycle disturbances and ego dissolution as the likely origins. PH has subsequently been associated with a range of differing central nervous system pathologies, including vascular and infectious midbrain [5], pontine [6] and thalamic lesions [7], local subarachnoid hemorrhage [8], compression by local [9,10] and distal tumors [11], basilar migraine [12], basilar vascular hy-

poplasia [13] and following regional surgical [14] or angiographic interventions [15].

The case we present has the peculiarity of having characteristics somewhat different from those described frequently in the cases published to date. One of the most special characteristics for which we have decided to publish it is the duration of the syndrome, until the date we have not found any other published case of superior duration. Our case does not criticize distorted perceptions of reality and understands them as part of his everyday life.

As a result, we will review the published literature and redefine the particular characteristics of this rare clinical picture.

Case Report

61-year-old male enters nephrology ward due to grade 3 renal failure and frequent falls during last month caused by "losing strength" in the legs. In addition, he presented a history of a thalamic brain accident evidenced by CT as a 4 mm lesion in the medial thalamus which had taken place 5 years ago and that had led to time-space disorientation, general discomfort, nausea and vomiting to the present. He had no history of personal or familiar previous psychiatric pathology.

Upon admission, the patient had no objectionable alterations in mental state, so the study and treatment of neuro and nephrological pathology began. The hydro electrolytic and acid base balance alterations were corrected and thoracic and cerebral abdominal imaging tests were performed trying to clarify "the loss of strength".

Head and neck MRI showed a hypoplasia of the A1 segment on the right Anterior Cerebral Artery and of the P1 on the right Posterior Cerebral Artery. To complete the study, we performed abdominal ultrasound, chest X-ray, pulmonary scintigraphy as well as doppler study of lower limbs that did not provide any relevant information. It was therefore determined that sudden episodes of falls were due to drops attacks: abrupt falls without prodromal symptoms trigger factors or loss of consciousness and with instantaneous recovery to baseline status.

Unexpectedly, during his stay in the hospital ward, the patient began to verbalize that for five years, just after suffering the thalamic infarction, he had begun to see images of his everyday life: "I saw my own son in the kitchen of the house eating a snack and did not understand why he did not offer me to seat and eat with

him." He described clear situations, vivid images with colors and movement and in which the subjects spoke to him, as if they were real. At first he experienced "anger and frustration, they were his son, his wife... but why they did not listen to me?" Eventually he learned to differentiate them from reality: "those characters spoke and acted ignoring him, they talked and moved around as if I wasn't there." A psychiatric evaluation of the patient was then carried out, who at all times had remained in a good state of consciousness and orientation. He referred a year evolution of visual and auditory hallucinations, currently identified as unreal but real until that moment and that did not create any kind of anguish or emotional repercussion.

Hallucinations occurred during the day and at night and were not associated with sleep disturbances, agitation or vision deficits. Attention, memory, language and cognition were intact (his Mini-Mental State Examination score was 30/30). There were no anxious or mood alterations, nor in the instincts. The patient verbalized fear of expressing these phenomena before that time "They would have taken me crazy!!" Therefore, a conservative treatment was carried out, explaining him the origin of these visions and reassuring him. Since they did not produce at the present time any type of repercussion at a behavioral or affective level, it was decided not to apply any pharmacological treatment.

A search in Pubmed was conducted on similar cases with the words "peduncullar hallucination or hallucinosis or psychosis and thalamic stroke or infarction or ischemia". Only 16 published reviews were found. In most cases there were multifocal lesions, including in most of them the midbrain and the hallucinations were of the "liliputian" type (See table 1).

We found a clear association of the condition with alterations in blood pressure in 6 of the 17 cases described in the form of malignant or uncontrolled hypertension due to organic cause or due to poor compliance with the treatment. Also in more than half of the cases there was a chronic kidney disease diagnosed of advanced degree and in almost all of them a concomitant diabetes mellitus. Delgado., et al. describe a case of hallucinations of thalamic origin but also with damage at the mesencephalic level in which the patient develops complex visual and auditory hallucinations, vivid and with a fantastic component (monsters, shadows...), to this type of hallucinations they call "disorteidolias" [16].

Only one case of those found had any type of previous psychopathology or were in follow-up in psychiatry [17].

Author	Age	Sex	Medical pa- thologies	Neurological Symptoms	Sleep dis- turbances	Peduncular damage	Visual hallucinosis	AH	Treat- ment	PD	Tox- ic	Dura- tion
Delgado., et al. 2013	61	М	Obesity	Paresthesias	No	Thalamic stroke (dor- somedial nucleous), sparing the intrala- minar nuclei and the midbrain	No	No	No	No	No	2 weeks
Speigel., et al. 2011	77	F	Hyperlipemia	Headache, diplopia and ptosis	No	Acute/subacute right midbrain infarct.	Lilliputian	No	Yes Olan- zapine	No	No	3 weeks
Mocellin., et al. 2006	85	F	Hypertension, Hypercho- lesterolemia, Diabetes	Visual impairment (15 years, glaucoma)	No	Multiple subcortical white matter lesion and bilateral lesions in the pons	Yellow and pink flowers, disembodied and grimacing male faces, mainly that of a James Bond actor appearing in the bodies of family members, uniformed children around the hospital bed	No	Yes Olan- zapine	No	No	2 days
	68	F	Hypertension, Hypercholester- olemia	Frontal Hemorrhage (38 years age)	No	Right thalamus lacu- nar infarct	Unusually small chil- dren dressed in colorful uniforms	No	Yes Risperi- done	No	No	3 weeks
	33	F	Arteriovenous malformation	Left tempo- roparietal ar- teriovenous malformation (embolized when 25 years age), migraine	No	Midtemporal epilep- tiform focus	Disembodied faces and torsos floating (moving their lips like talking to her), red letters on people's faces and her own face in the mirror, hundreds of little spikes and small ants moving (predominantly in the right visual field),	No	Yes Phen- itoin	No	No	1 week
Danziger., et al. 1997	56	M	Aortic stenosis	No		Unilateral infarct restricted to the cerebellum, rostral protuberance and posterior thalamus.	n.e	No	No	No	No	1 day
Serra Catafau., <i>et</i> <i>al</i> . 1992	68	М	Unknown	Hemiparesis, paraesthesias on the left side	No	Right posterior tha- lamic infarct as the sole lesion.	n.e	No	No	No	No	5 days
Feinberg., et al. 1989	83	М	Diabetes	No		MRI: Paramedial right thalamic infarc- tion without anor- mality of cerebral pe- duncles or midbrain	n.e	No	No	No	No	3 days
Kölmel, 1990	56	M	Heart disease	Diplopia	Yes	Bilateral ischemic lesion in thalamus and mesencephalon	n.e	Yes	Yes (n.e)	No	No	>24 hours

Mollet., et al. 2007	61	F	Hyperlipidemia, morbid obesity, hypothyroid- ism, hyperten- sion, coronary artery disease, and diabetic peripheral neuropathy		No	Right thalamic stroke	College boys	Yes	No	Yes	No	n.e
Fornaz- zari., <i>et al</i> . 2012	45	M	Hypertension	Memory impairment	No	Left lateral poste- rior nucleus of the thalamus	Sound-tactile, sound- color, and grapheme- gustatory synesthesia	No	No	No	Yes	9 months
Badrin., et al. 2017	73	M	Diabetes and hypertension	No	Yes	Right thalamus	n.e	Yes	Yes Risperi- done, ISRS	No	No	Un- known
Liao., <i>et al</i> . 2018	67	F	Hypertension, hyperlipidemia	Ataxia, dys- arthria, scan- ning speech	No	Bilateral and cerebel- lum	Persecutory delusions	No	Halo- peridol	No	No	2 months
Santos., et al. 2009	49	M	Lymphoblastic leukemia	No	No	Left thalamus	No. Gustative hallucina- tion	No	Yes que- tiapine	No	Alco- hol	3 months
Crail Melendez., et al. 2013	37	F	Unknown	No	No	Posterior region of the right thalamus	Persecutory delusions	No	Yes, Risperi- done	No	To- bac- co	1 week
Mittal., et al. 2010	19	F	Factor V Leiden mutation	No	No	Left thalamus	Ex boyfriend	No	Yes Risperi- done	No	No	1 month
McGil- christ., et al. 1993	43	M	Unknown	No	Yes	Bilateral thalamic infarction	Delusions of self-reference and grandiosity and visual hallucinations.	No	Yes Lithium	No	No	18 months
Arikan., et al. 2009	33	M	Unknown	Speaking, learning and concentration difficulties	No	Left thalamic lacunar infarcts	Auditory hallucinations and delusions of perse- cution	No	Yes, Risperi- done	No	No	2 weeks

Table 1: Articles that includes case reports of peduncular hallucinosis and thalamic stroke.

Note: F: Female; M: Male; n.e: Not Specified; AH: Auditory Hallucinosis; PD: Psychiatric Disorder.

In a majority of the reviewed cases it is necessary to administer some type of pharmacological treatment, especially if the symptoms are accompanied by behavioral or personality changes. Risperidone is the antipsychotic that has been most frequently used in those patients in whom pharmacological treatment has been necessary [18-21]. However we found a published case in which the use of olanzapine quetiapine at low doses or even lithium had a satisfactory effect for their control [18,22,23].

Synesthesias were reported in two of the cases analyzed [22,24].

As in our case Kolmel., *et al.* also described a single thalamic lesion on the CT scan and hallucinations are also indistinguishable. Another differential factor was that despite how vivid they were in both cases, indistinguishable from reality, as in our patient, in the published case corresponded with images of the past and included people who had died years ago.

The main differences with the case we presented was the duration of them, about five years, with a decrease in their intensity and complexity afterwards without the need for specific treatment and which were accompanied by auditory hallucinations.

Also the origin is a unique lesion in the thalamus region not associated with other peripheral ischemic lesions.

Conclusion

We present an updated review of the literature published to date on thalamic infarction accompanied by psychotic symptomatology. Mainly we find descriptions of classic cases and only two cases published in the last 5 years. We analyze the common and differential characteristics of each of them in order to understand and redefine the concept of peduncular hallucinations secondary to thalamic ischemic events.

The classic descriptions of authors such as Feinberg in 1989, present data of a patient with a pyramidal right thalamic infarction that presented vivid hallucinations. Serra Catafau in 1992, describes visual hallucinations in a patient with a posterior thalamic infarction.

In the first description of Lhermitte, they appear as associated with sleep disorders, although we only found that association in three of the cases we have analyzed [1,19,22].

Although the exact pathogenesis of peduncular hallucinations is still unknown, in most cases it affects the midbrain or thalamus, and in the majority of published cases it appears after a heart attack that damages these structures. Half of the cases there are some other neurological deficits, like paresthesias [16] diplopia and ptosis [21,25] ataxia or dysarthria [20,26]. Nor are there other alterations in the higher cognitive functions of attention, concentration, orientation, consciousness or intellect. There are no other accompanying mental symptoms that suggest psychopathological alterations.

Because sleep disturbances often accompanied the visual hallucinations, Lhermitte believed that peduncular hallucinosis resulted from the pathological release of subcortical regions that are active in dreaming, while consciousness remained intact. In addition to lesions in the midbrain, injury to the pulvinar nucleus of the thalamus and lower pons also has been associated with complex visual hallucinations, raising the possibility that this condition may arise

from disruption of subcortical visual processing pathways or a diffuse cerebral reaction to various lesions in primitive structures.

A significant overlaps exists between the concepts of Charles Bonnet Syndrome and PH. The fluidity of these eponymous syndromes reduces their validity and meaning, and may result in an inappropriate attribution of the underlying pathology. Charles Bonnet Syndrome describes a group of patients with visual hallucinations in the setting of visual loss, and shows significant clinical overlap with PH. An understanding of how differing pathologies may produce complex visual hallucinosis allows the appropriate understanding of treatment, depending on the site and nature of the lesion and content of perceptual disturbance [18] (See table 2). According the literature reviewed so far, we understand that an exhaustive ophthalmological study of patients would be recommended in this type of case to make a differential diagnosis with Charles Bonnet Syndrome [27,28].

Peduncular hallucinosis	Charles Bonnet syndrome				
Percepts predominantly visual	Percepts predominantly visual				
Percepts vivid, formed and colorful	Percepts vivid, formed and color- ful				
May include Lilliputian phenomena	May include Lilliputian phenom- ena				
Stimuli animate, mobile and non-stereotypical	Varied				
Predominate at night	Predominate at night				
Generally clear sensorium	Generally clear sensorium				
May have ocular disease; Central Nervous System pa- thology frequently reported	Frequently have ocular disease; Central Nervous System pathol- ogy rarely reported				
Occur across age groups	Most common in elderly				

Table 2: Comparative features of peduncular hallucinosis and Charles Bonnet syndrome.

Regarding treatment in most cases it has been decided not to use antipsychotic treatment because the individuals did not describe them as bothersome and the side effects of the medication in this type of generally aged population could be more harmful.

Therefore, within hallucinatory cases of an organic type, especially in patients with cardiovascular risk factors, it is important to rule out rare syndromes such as Charles Bonnet or hallucinations

of thalamic origin, being always indicated to perform brain and ophthalmologic imaging tests.

We present, therefore, what we believe is one of the only cases in which a picture of hallucinosis is presented as the origin of a single lesion at thalamus level and the extension of the complex has been prolonged than any case described to date.

It is noteworthy the degree of reality and familiarity that has made our patient live with their symptoms for five years, the longest of the many cases reviewed in the existing bibliography, duration that would include the status designation.

Conflict of Interest

The authors report no conflict of interest.

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