Auditory Hallucinosis as a Presenting Feature of Interpeduncular Lipoma with Proximal P1 Segment Fenestration: Report of a Rare Case and Review of Literature on Peduncular Hallucinosis

Ashish Kulhari, MD,1 Sunil Manjila, MD,2 Gagandeep Singh, MD,3 Kunal Kumar, MD,1 Robert W Tarr, MD,1 and Nicholas Bambakidis, MD2

1 Department of Neurology, Neurological Institute, University Hospitals Case Medical Center, Cleveland, OH, USA
2 Department of Neurological Surgery, Neurological Institute, University Hospitals Case Medical Center, Cleveland, OH, USA
3 Department of Neuroradiology, Neurological Institute, University Hospitals Case Medical Center, Cleveland, OH, USA

Abstract

The authors present a unique case of intracranial lipoma in the interpeduncular cistern associated with proximal P1 segment fenestration. This patient is a 20-year-old male with extensive psychiatric history and complaints of recent auditory hallucinations. Cranial magnetic resonance imaging (MRI) (T1, T2, and FLAIR) showed a hyperintense lesion in the left aspect of interpeduncular cistern with a prominent flow void within the hyperintense lesion suggestive of a combined vascular–lipomatous lesion. Computed tomography (CT) angiography showed a high-riding large tortuous P1 segment on the left side with proximal fenestration, the ectatic posteromedial limb harboring a fusiform dilated segment. Since there are anecdotal cases of cerebral aneurysms associated with intracranial lipomas, a conventional angiography was done, which confirmed a proximal left P1 fenestration and a fusiform-dilated segment, and no aneurysm. There are few cases of hallucinations associated with a vascular midbrain pathology reported in literature, but hallucinations associated with a combination of lipoma and arterial ectasia have never been reported. This article not only demonstrates the MRI and angiographic appearance of this rare lipomatous lesion but also highlights this unique association and significance of auditory hallucinations as a clinical presentation, akin to peduncular hallucinosis.

Keywords

Auditory hallucinosis; interpeduncular lipoma; peduncular hallucinosis; posterior cerebral artery fenestration; vascular ectasia

INTRODUCTION

Intracranial lipomas are rarely occurring congenital nervous tissue malformations that contribute to 0.1% of all intracranial tumors. They arise from abnormal differentiation of mesenchymal tissue of meninx primitive [1,2]. The most common location for these benign neoplasms is at or near the midline. These lesions are found most frequently in the pericallosal–interhemispheric region (45%), followed by the quadrigeminal cistern (25%), and the suprasellar/interpeduncular cistern (14%). Intracranial lipomas are mostly asymptomatic and are found incidentally on brain imaging. When symptomatic, they commonly cause headache and seizures. Rarely, these lesions can cause obstructive hydrocephalus. We report a unique case of interpeduncular lipoma, associated with arterial fenestration of the left P1 segment, masquerading as a likely aneurysm and presenting with visuo-auditory hallucinations.

Peduncular hallucinosis (PH), known as Lhermitte’s peduncular hallucinosis, was originally described by French neurologist J. Lhermitte in 1922. Later, it was termed “peduncular hallucinations” by Von Bogaert [3–5]. It is a rare neurological disorder, which is characterized with vivid colorful visual-acoustic hallucinations that typically occur in a dark environment, last for several minutes [6–8] and are composed of moving patterns, people or animals [9,10]. Unlike some other types of hallucinations, the hallucinatory experiences of PH...
appear to be very realistic. It has been reported that peduncular hallucinations may occur due to infarction, vasospasm, or compression of intracranial structures such as thalamus, pars reticulata of substantia nigra, midbrain, pons, and basal diencephalon [11–14].

ILLUSTRATIVE CASE

This 20-year-old male with past history of depression was admitted to our institution with new onset visual hallucination. He had an extensive psychiatric history with physical and sexual abuse, auditory hallucinations since childhood, suicide attempt at the age of 14, poly-substance abuse in the recent past and occasional alcohol consumption. He had been on sertraline since the age of 18, which he stopped taking as it was not helping him. At the age of 6, he started hearing voices mostly when he was alone. These voices were initially that of a single male, repeatedly mumbling to him, without any suicidal or homicidal content, not really bothering him or affecting activities of daily living. For the past year, he was hearing two voices, talking with each other, with suicidal and negative content. These auditory hallucinations had considerably increased in frequency and intensity. Sometimes he saw poorly defined images, objects, and shadows, which he was unable to identify yet knew were unreal. Vital signs and physical examination including neurologic examination were normal. Initial laboratory investigation including routine hemogram, metabolic panel, drug screen, thyroid profile, Vitamin B12, and folate levels, HIV and syphilis antibody were unremarkable. Noncontrast CT head showed hypodense lesion in the left ventral perimesencephalic region with no adjacent edema or significant mass effect (Fig. 1).

Cranial MR imaging showed a single extra axial hyperintense lesion in the left aspect of interpeduncular space on T1 and T2 sequence suggestive of fatty tissue (Fig. 2).

There was a central prominent flow-void within this hyperintense lesion, which probably represented a tortuous left P1 segment. The differential diagnosis of an interpeduncular lipoma versus interpeduncular bleed was entertained. CT angiography demonstrated a high-riding large tortuous P1 segment on the left side with a proximal fenestration where the ectatic posteromedial limb had a fusiform dilatation, possibly aneurysm (Fig. 3).

Confirmatory conventional catheter-based cerebral angiogram showed a high riding large tortuous P1 segment on the left side with a proximal fenestration where the ectatic posteromedial limb has a fusiform dilatation; no evidence of a distinct aneurysm was seen (Fig. 4).

At this time, the final diagnosis of an interpeduncular lipoma associated with an ectatic fenestrated P1 segment was made. Given the extensive psychiatric history, we treated him with antipsychotics. Unfortunately, the patient was lost to follow-up with us. There are only a handful of case reports on hallucinations associated with a vascular midbrain pathology, while that associated with a lipoma and vessel ectasia are never reported.

DISCUSSION

Intracranial lipomas develop from the abnormal persistence and mal-differentiation of the mesenchymal remnants of meninx primitiva as proposed by Verga et al [2] and later supported by Truwit and Barkovich [1]. Most reports in the literature suggest that intracranial lipomas arise in the pericallosal or dorsal mesencephalic cisterns and Sylvian fissure [15,16]. The interpeduncular, quadrigeminal cistern and perimesencephalic lipomas are relatively less common [16]. Usually, midline lipomas are associated with vascular anomalies like aneurysms and are mostly asymptomatic, while lateral lipomas like those located in the Sylvian fissure typically present with seizures. Lipomas of the corpus callosum associated with distal ACA aneurysms and those of the quadrigeminal plate associated with communicating artery
Figure 4: a, b, c and d. Conventional cerebral angiogram confirmed a high riding large tortuous P1 segment on the left side with a proximal fenestration better seen in (d) where the ectatic posteromedial limb has a fusiform dilatation; no evidence of a distinct aneurysm was seen.

Aneurysms are reported respectively in the literature [17–20]. Interpeduncular fossa lipoma can compress the oculomotor nerve palsy [21]. Similarly, dorsal mesencephalic lipoma has been reported to be associated with inferior collicular hypoplasia [22]. Similar to arterial abnormalities associated with intracranial lipomas, venous abnormalities like sagittal sinus and falcine sinus fenestrations are reported by Ahmetogu et al [23].

Our patient presented with a significant worsening of auditory hallucinosis and new onset of ill-defined visual hallucinations and was found to have fenestration of the
P1 ectatic segment adjacent to the lipoma. Considering the midbrain compression, the clinical diagnosis of "peduncular hallucinosis" was entertained—a syndrome of naturalistic, complex, scenic, mostly visual, or visual–acoustic hallucinations that recurs stereotypically over months [4,24]. Unlike the hallucinations seen in psychosis, peduncular hallucinations are not usually bizarre and usually involve people and environments that are familiar to the affected individual. Because of the nonbizarre nature of peduncular hallucinations, these hallucinations seem very real to patients [24]. Unlike psychosis, PH patients are aware of the fact that these hallucinations are not real. In addition to hallucinations, PH patients may present with confusion, abnormal behavior, delusional misidentification for persons and places, oculomotor disturbances, ataxia, dysarthria and loss of disease awareness. In the chronic stages of PH, the sleep-wake cycle may get disturbed [24]. Patients with PH often have severe impairment of episodic memory, occasionally coupled with confabulatory behavior [25].

PH has been reported in cases of vascular and infected lesions of the thalamus, the pars reticulata of substantia nigra, midbrain, pons, and basal diencephalon [11,26–31]. PH have also been reported in cases of ruptured basilar-superior cerebellar artery aneurysm, vertebral angiography, and transient brain stem compression [32–34]. It is also more common in patients with a long duration of Parkinson’s disease, depression, and cognitive impairment [35]. The physiology of hallucinations in PH is not well understood. Spiegel et al postulated “excessive excitatory cholinergic activity at lateral geniculate nucleus due to loss of inhibitory ascending serotonergic pathway from pons” as the possible physiology of PH [8]. The clinical phenomenology of peduncular hallucinations overlaps with those hallucinations that are observed in patients with severe visual impairment like that found in Charles Bonnet syndrome. It appears that the term "peduncular hallucinosis" might beneficially be reserved only for cases with documented mesencephalic and/or thalamic involvement and in whom one can confidently rule out a temporal association with neurological involvement in other regions of the brain (e.g., in patients with visual pathway lesions or epileptic hallucinations), ophthalmological disorders including Charles Bonnet syndrome, delirium, or psychopathology.

Neurobiology of PH is not well understood. As cited before, Spiegel et al postulated excessive unopposed excitatory cholinergic activity at lateral geniculate nucleus due to loss of inhibitory ascending serotonergic pathway from pons as the possible physiology of PH [8]. Because PH is thought to be due to loss of inhibitory action of serotonergic neurons, atypical antipsychotics, like olanzapine and quetiapine, which block both dopamine and serotonin receptors and increase the amount of serotonin in the brain have shown to be effective in treatment of PH [8,35]. Selective serotonin reuptake inhibitors like citalopram have also shown to be beneficial in treating PH [36]. Anticonvulsants, like carbamazepine, which increase the central GABA activity and inhibit LGN from firing have shown to be useful in controlling peduncular hallucinations [37]. Sedatives and other CNS suppressants like alcohol and benzodiazepines can decrease the threshold of any type of hallucination and so it is useful to decrease intake of these substances. Psychotherapy and family support has also shown to be useful in PH.

Surgical lesions associated with PH reported in the literature include ruptured basilar-superior cerebellar artery aneurysm [29], brainstem compression by cerebellar metastasis [34] or pineocytoma [37], and primary cerebellar tumors like pilocytic astrocytoma and medulloblastoma [30]. Surgical resection of these lesions has proven to be useful in reducing PH [37]. In short, this intriguing clinical syndrome should warrant adequate intracranial screening, including appropriate vascular imaging of the brain. The importance of associated vascular lesions with intracranial lipomas cannot be overemphasized.

REFERENCES