BEAST ON HER SHOULDER: A RARE PRESENTATION OF PEDUNCULAR VISUAL HALLUCINATION IN A CASE OF CRANIOPHARYNGIOMA

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INTRODUCTION

Craniopharyngioma is a rare, benign, and slow-growing brain tumor. It is typically located over the sellar and suprasellar region, constituting 1%-5% of primary brain tumors with low incidence rate of one per million person-years approximately (Bunin et al.1998). Headache, raised intracranial pressure, and hydrocephalus are common issues related to mass effect. Some patients manifest visual field defect and endocrinal disturbance due to the anatomical proximity to the optic chiasma and hypothalamic-pituitary axis, respectively (Muller et al. 2019). Till date, only few cases of psychopathological symptoms related to craniopharyngioma have been reported. Herein, we present the case of a patient who experienced visual hallucinations and persecutory delusion. Our findings indicate that slow-growing brain tumors might occasionally present primarily as neuropsychiatric condition and tumour is diagnosed during further case work up.

CASE HISTORY

A 55-year-old female graduate with no psychiatric or medical illness in the past as well as in family, presented to us with an illness for 12-month duration. It was an insidious onset, continuous course with symptoms initially characterized by frontal headache not associated with nausea vomiting or blurring of vision from 1year which was gradually increasing, visual hallucinations (seeing a lady on a lion which will scare the patient), from 10 days. She would frequently complaints of these hallucination to family members from last 10 days, which initially family members attributed to some evil spirits, but as it persisted they brought her to psychiatry OPD for further treatment There was no history of swallowing or speech problems, abnormal eye movements, apraxia, agnosia, paraesthesia, chronic exposure to any toxin, recent infection or fever, substance abuse, excessive anxiety, or obsessive-compulsive symptoms.

On examination, her BP was 130/80 mm of mercury and pulse rate was 84/min. General physical examination was within normal limits and systemic examination revealed no abnormality. A detailed neurological examination was done which revealed sluggish pupillary reflexes along with decreased visual acuity on both side. Motor and sensory examination was found to within normal limit. Ophthalmological examination revealed slight optic disc oedema with sluggish pupillary reflexes in bilateral eyes. Visual acuity was decreased to 6/36 in both eye. MRI brain with contrast was done suspecting intracranial pathology. Magnetic resonance imaging (MRI) revealed well-defined T1-hypointense and T2-heterogenous hyperintense solid cystic lesion in suprasellar location measuring 3.7 cm \times 2.7 cm \times 2.1 cm; intensely heterogeneously enhancing internal solid component with peripheral cystic component demonstrating thin, smooth peripheral rim enhancement. No diffusion restriction was noted within the lesion. Other investigations, such as hematology, renal and liver function tests, thyroid function tests, chest X-ray, blood culture, urine routine, and microscopy, were within normal limit. Provisional diagnosis of craniopharyngioma with visual hallucinations was made. For visual hallucinations and behavioural disturbances patient was started of Tablet risperidone 3mg/day. Neurosurgical opinion was taken, and they opined for surgery to be planned in next 15 days. In meanwhile on medication, patient improved significantly. Patient would complaint about visual hallucination less often than before and sleep was also improved.

DISCUSSION

Craniopharyngioma (CP), remnants of the embryonic Rathke's pouch, is among the most frequently reported tumors that can manifest in both childhood (most common) and adulthood, usually involving the posterior hypothalamus. CPs presenting as incidental findings are rare (<2% of all CP cases) (Boekhoff et al. 2007). Most

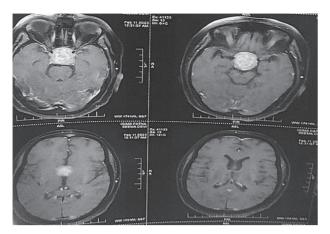


Fig 1. (MRI) imaging showing well-defined T1hypointense and T2-heterogenous hyperintense solid cystic lesion in suprasellar location

common manifestations are endocrine deficits (52-87% of patients) and visual impairments (62-84% of patients) (Prieto et al. 2015). The type and degree of visual impairment depend on the anatomical tumour topography with regard to optic chiasma distortion. As most are suprasellar, the patients usually present with visual abnormalities and headaches. As these tumours are often slow growing, many times patient being diagnosed as incidental finding is not that uncommon. In our case, although the patient was having headache from 1 year, the visual hallucinations were the presenting complaint. Peduncular hallucination as described as early as 1952 were vivid, coloured visual images of people, animals, plants, scenes, or geometric patterns (Rozanski 1952). Peduncular hallucinations have been associated with brainstem lesions of vascular and infectious etiology. Cogan divided visual hallucinations into release and irritative phenomena (Cogan 1973). Irritative phenomena are paroxysmal and stereotyped hallucinations that may assist in localization of a lesion. An occipital lobe focus produces lights or colors, whereas a temporal lobe focus results in complex formed image. Release phenomena are continuous hallucinations in which the images may change from one episode to the next. They are attributed to complete or partial absence of normal visual input, occur with lesions at any level of the visual system, and do not localize lesion. Peduncular hallucinations are a form of release phenomena. Disturbances of sleep-wake cycles are often associated and may help in recognizing peduncular hallucinations.

Various case reports of craniopharyngiomas presented with altered mental states, including a rapidly progressive dementia, mania, catatonia and psychosis. Psychiatric symptoms occur in craniopharyngiomas due to the lesion disrupting neural connections between the hypothalamus and the limbic system (Bowers & Hughes 2021). The absence of neurological signs in this case is consistent with a study of 148 patients with craniopharyngeoma in which only 47% had visual disturbances; other neurological symptoms were relatively uncommon (Fahlbusch et al. 1999). The abnormal cognition seen in this patient is common with pituitary tumors. New learning and recent memory are often impaired, while remote memory and other cognitive functions are usually normal (Shin et al. 1999). The neurobehavioral changes in pituitary tumors may be secondary to endocrine abnormalities or to compression of structures surrounding the sella. Before brain imaging was not readily available, up to 70% of cases of 'psychosis due to brain tumor'were missed, especially if neurological symptoms were absent (Sinai et al. 2003).

In our case, mostly pressure from tumour might be leading to release type peduncular hallucinations, which got drastically improved with antipsychotic medications cover along with supportive measures provided by neurosurgery consultation targeting decrease in intracranial pressure due to mass.

To conclude, visual hallucination in this case was a boon in disguise because it paved the way for a diagnosis of craniopharyngioma. We propose a comprehensive and holistic workup in late middle-aged women with sudden onset visual hallucination in the background of medical symptoms. We should always be vigilant for craniopharyngioma as aetiology for rare presentation of visual hallucination.

Ethical Considerations: Does this study include human subjects? YES

Authors confirmed the compliance with all relevant ethical regulations.

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References

- Boekhoff S, Bison B, Eveslage M, Sowithayasakul P & Müller HL: Craniopharyngiomas presenting as incidentalomas: results of KRANIOPHARYNGEOM 2007. Pituitary 2019;22:532-41.
- 2. Bowers AR & Hughes L: Craniopharyngioma presenting with psychiatric symptoms: a case report. Prog. Neurol. Psychiatry 2021;25:18-20.
- 3. Bunin GR, Surawicz TS, Witman PA, Preston-Martin S, Davis F, Bruner JM: The descriptive epidemiology of craniopharyngioma. J Neurosurg. 1998;89:547–51.
- 4. Cogan DG: Visual hallucinations as release phenomena. Albrecht Von Graefes Arch. Klin. Exp. Ophthalmol 1973;188:139-50.
- Fahlbusch R, Honegger J, Paulus W, Huk W, Buchfelder M: Surgical treatment of craniopharyngiomas: experience with 168 patients. J. Neurosurg 1999; 90:237-50.

- 6. Muller HL, Merchant TE, Warmuth-Metz M, Martinez-Barbera JP, Puget S: Craniopharyngioma. Nat Rev Dis Primers 2019;5:75. doi: 10.1038/s41572-019-0125-9
- 7. Prieto R, Pascual JM, Barrios L: Optic chiasm distortions caused by craniopharyngiomas: clinical and magnetic resonance imaging correlation and influence on visual outcome. World neurosurg. 2015;83:500-29.
- 8. Rozanski J: Peduncular hallucinosis following vertebral angiography. Neurology 1952;2:341-9.
- Shin JL, Asa SL, Woodhouse LJ, Smyth HS, Ezzat S: Cystic lesions of the pituitary: clinicopathological features distinguishing craniopharyngioma, Rathke's cleft cyst, and arachnoid cyst. J. Clin. Endocrinol. Metab. 1999;84:3972-82.
- 10. Sinai J, Wong AH: Craniopharyngeoma presenting as psychosis, disinhibition and personality change without neurological signs. Acta Neuropsychiatr. 2003;15:94-6.

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