

CASE REPORT

Spontaneous, isolated, and gaze-evoked ocular flutter: A rare case report

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Abstract

This case illustrates the need to correctly identify oscillopsia among cases of self-reported dizziness, and ocular flutter itself as a presenting sign of potentially serious underlying disease. The neurologist should approach these patients in a systematic manner, in order to exclude concurrent or impending comorbidities such as neoplastic disease.

KEYWORDS

ocular flutter, ocular motility, oscillopsia

1 | INTRODUCTION

Ocular flutter is a rare symptom in the context of many diseases. We present a case with ocular flutter that resolved completely in 2 weeks. To our knowledge, this is the first report of an adult patient with isolated, gaze-evoked, ocular flutter without any significant concomitant disease or precedent infection.

Ocular flutter is a rare disorder characterized by pathological, involuntary eye movements, occurring strictly in the horizontal plane as conjugate horizontal saccades; in opsoclonus, the eye movements could be in any plane (both horizontal and vertical).¹ The true prevalence of the syndrome may be obscured by self-limiting cases. Ocular flutter is a rare nonmetastatic symptom that may exist within the context of underlying malignancies (neuroblastoma in children, breast and lung cancer in adults, and ovarian teratoma), as paraneoplastic phenomenon or in the context of

autoimmune diseases (eg, multiple sclerosis). Rare causes of ocular flutter could exist due to drug intoxication (eg, lithium, cocaine, and phenytoin) or chemical poisoning (eg, organophosphates and toluene) and head trauma. Rarely, ocular flutter appears as a postviral syndrome (postviral encephalitis), or deemed “isolated,” when it cannot be attributed to any identifiable cause.^{2,3}

2 | CASE PRESENTATION

A 34-year-old woman presented to outpatient clinic with acute onset intermittent oscillopsia. Her medical history was unremarkable. Physical examination was normal. The neurological examination revealed eye oscillations with preponderance on the horizontal plane (ocular flutter), evoked by gaze fixation regardless of gaze direction and supraversion (Video S1). She did not report diplopia, and there were no

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other remarkable findings. Brain magnetic resonance imaging (MRI) before and after paramagnetic medium was unremarkable.

Whole body computerized tomography (CT) without and with contrast medium was performed on the premises of a potentially undiagnosed neoplasm; it was, however, without any abnormal findings. Following a lumbar puncture, cerebrospinal fluid (CSF) cytology and biochemistry were equally within normal range and showed lack of acute inflammation, both viral and immunological (absence of oligoclonal bands, negative cultures, and polymerase chain reactions for viral, bacterial, and fungal infections). Opening pressure of the CSF was within the normal range. An intensive workup for serum ganglioside and paraneoplastic antibodies was also negative. The symptom itself resolved completely within 2 weeks from its onset, without any therapy. One year later, the neurological examination of the patient was normal, as well as all repeated paraclinical investigations (paraneoplastic antibodies and CT examinations). Our intention is to continue following this patient and repeat the examinations (clinical and paraclinical) at the follow-up appointments.

3 | DISCUSSION

To our knowledge, this is the first report of a patient with an adult-onset, gaze-evoked, isolated ocular flutter without any significant concomitant disease or infectious precedent. Literature search in PubMed using the terms “Isolated ocular flutter” retrieves ten articles, out of which only two describe adult-onset cases,^{2,3} indicating that it is a rare syndrome though the possibility of its underdiagnosis cannot be excluded; the main reasons for this could be that the symptomatic presentation itself (ie, oscillopsia) may be mild and with rapid resolution. Ocular flutter as the only preceding symptom of an underlying malignancy is also very rare. Some cases presented with isolated ocular flutter and the ensuing workup added with an early lung adenocarcinoma that would not have been found.^{4,5}

Association of ocular flutter with generalized myoclonus, trunk ataxia, and positive ganglioside antibodies points to a possible autoimmune pathology of the syndrome.⁶ Demyelinating pontine lesions could interrupt the tonic stimulation of omnipause neurons to burst neurons within the paramedian pontine reticular formation (PPRF) which is responsible for conjugate horizontal eye movements leading to ocular flutter, as has been described in a case of multiple sclerosis.⁷ Brain MRI imaging does not always demonstrate brainstem lesions, as in our case, suggesting a functional rather than a structural central nervous system (CNS) dysfunction. Circulating antibodies might target CNS epitopes within the PPRF.⁶ Moreover, small lesions in the PPRF could be missed on 1.5 or lower Tesla MRIs. Therefore, it should

be worthy to perform MRIs in higher than 1.5 Tesla in order to increase the possibility to detect small lesions, such as demyelinating lesions, within the PPRF area. Spontaneous remission of ocular flutter has been also described in reported cases, again strengthening the origin of autoimmune hypothesis of this rare syndrome.⁶

4 | CONCLUSIONS

Ocular flutter and ocular motor disorders in general, although rare, represent an important finding given their integration in paraneoplastic syndromes; as such, despite its potential for partial or full resolution, increased vigilance is warranted by the physicians for signs of latent systemic disease or occult malignancies.

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CONFLICT OF INTEREST

Authors declare no conflicts of interest.

AUTHORS' CONTRIBUTION

All authors: contributed equally to the manuscript and accepted the final version of the manuscript.

ETHICAL STATEMENT

Following a detailed explanation of the procedures involved and the use of de-identified data for the purposes of this manuscript, the patient provided informed consent on the use of said de-identified data, including the relevant video.

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