CASE REPORT

Ophthalmoplegic migraine with isolated third cranial nerve palsy in a known case of juvenile myoclonic epilepsy

Aakash Shetty,¹ Supriya Khardenavis,² Anirudda Deshpande¹

¹Department of Neurology, Kasturba Medical College, Manipal, Manipal, Karnataka, India

²Department of Ophthalmology, JJM Medical College, Davanagere, Karnataka, India

Correspondence to

Dr Anirudda Deshpande, dr.aniruddha.deshpande@gmail.com

SUMMARY

Among the various forms of migraine headaches, ophthalmoplegic migraine is an uncommon and rare form, the incidence of which is approximately 0.7 per million. It presents predominantly with headache and ophthalmoplegia. One of more cranial nerves can be affected, however the third cranial nerve is most often affected. As a result, symptoms wise, mydriasis and ptosis are commonly seen. Patients generally recover completely within a few days or weeks, however residual deficits are known to occur in a minority of patients. One of the common generalised epilepsy syndromes is the juvenile myoclonic epilepsy (JME), its prevalence being roughly up to 10% of all patients with epilepsy. It usually begins in the second decade of life. Generalised tonic-clonic seizures myoclonic jerks absences constitute the main seizure types in JME. Studies indicate a definite association of epilepsy with migraine headaches and a significant number of migraineurs are found to be epileptic. Conversely, patients with epilepsy are two times more likely to have migraine, as compared to their first degree relatives without migraine. We report a known case of a female patient of JME having a history of classical migraine with aura presenting to us with headache and ophthalmoplegia. She was extensively evaluated to rule out other causes of isolated third cranial nerve palsy, with all the investigations being negative for any obvious cause. She was treated with non-steroidal anti-inflammatory drugs for the acute attack and was subsequently put on antimigraine medication, propranolol during her hospital stay, with which her ptosis recovered completely after 2 weeks. The patient was later started on tablet divalproex sodium, which the patient continues to take on a long-term basis, especially because of its efficacy as an antimigraine prophylaxis agent and a potent drug against JME.

BACKGROUND

Ophthalmoplegic migraine is a rare form of migraine headaches. It is even rarer to have a combination of isolated third cranial nerve palsy, all occurring in a patient of juvenile myoclonic epilepsy.¹⁻¹²

Although the association of migraine in patients with epilepsy and vice versa has been mentioned in medical literature frequently, it is rare to have the combination of isolated third cranial nerve palsy in ophthalmoplegic migraine in a patient of juvenile myoclonic epilepsy.

CASE PRESENTATION

A 26-year-old woman presented with a history of migraine headaches with aura since 10 years, and seizure disorder since 8 years, where in she used to report of things suddenly dropping out of her hands, especially in the mornings after getting up from sleep in addition to episodes suggestive of generalised tonic-clonic seizures. The patient was on tablet valproate sodium for the same since the past 2 years, with which there were no further episodes.

The patient developed ongoing severe left-sided hemicranial and throbbing headache (both starting almost simultaneously) for 3 days since the acute onset of ptosis of the left eye 3 days before coming. This was a second such attack on the same side as the previous attack a year ago. There was no documentation available of the previous such episode.

On examination, the left medial rectus was mildly restricted, the pupil was 3.5 mm reactive on the left side as compared to the right side 3 mm. Funduscopy was normal. There were no signs of meningeal irritation. No other cranial nerve involvement on examination was found. Rest of the neurological examination was normal.

Based on the history and examination, we thought of the following differential diagnosis as (1) ophthalmoplegic migraine, as historically and clinically it abided by the diagnostic criteria for ophthalmoplegic migraine, International Headache Society, international classification of headache disorders-second classification. Some of the other differentials we wanted to rule out were, (2) left posterior communicating artery aneurysm, (3) Tolosa-Hunt syndrome, (4) possible chronic meningitis, (5) myasthenia gravis and (6) congenital paralysis of levator palpebrae superioris. We investigated the patient through MRI of the brain with contrast, cerebrospinal fluid (CSF) analysis, neostigmine test and repetitive nerve stimulation; and other laboratory investigations such as antinuclear antibody (ANA), antineutrophil cytoplasmic antibody (ANCA). All of the aforementioned investigations were negative.

The patient was started on non-steroidal anti-inflammatory drug (NSAID) naproxen 500 mg twice daily and antimigraine medication in the form of β -blockers (propranolol) was given during her hospital stay. There was complete recovery of ptosis of the left eye in 2 weeks.

The patient was later started on tablet divalproex sodium, which the patient continues to take on a

CrossMark

To cite: Shetty A, Khardenavis S, Deshpande A. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-201718

long-term basis, especially because of its efficacy as an antimigraine prophylaxis agent and potent drug against juvenile myoclonic epilepsy.

The image collage depicts cardinal extraocular movements in every direction with normal range, except restricted left medial rectus (left eye adduction; figure 1).

INVESTIGATIONS

Complete blood count with erythrocyte sedimentation rate and C reactive protein were negative; MRI of the brain with contrast—normally no enhancement was seen; CSF analysis was normal; neostigmine test—no improvement of ptosis; and repetitive nerve stimulation—no decremental response. Other laboratory investigations like ANA, ANCA were negative.

DIFFERENTIAL DIAGNOSIS

- ▶ Ophthalmoplegic migraine
- ▶ Left posterior communicating artery aneurysm
- ▶ Tolosa-Hunt syndrome
- ▶ Possible chronic meningitis
- ▶ Myasthenia gravis
- ▶ Congenital paralysis of the levator palpebrae superioris

TREATMENT

The patient was started on NSAID naproxen 500 mg twice daily, antimigraine medication in the form of β -blockers (propranolol) 40 mg 1-1-0 was also started during the patient's hospital stay and she was put on divalproex sodium, as a long-term

antimigraine medication especially due to its high efficacy against juvenile myoclonic epilepsy.

OUTCOME AND FOLLOW-UP

There was complete recovery of ptosis of the left eye in 2 weeks. The patient was put on divalproex sodium, as a common, long-term antimigraine and antiepileptic medication. No similar attack has been reported after discharge.

DISCUSSION

Ophthalmoplegic migraine is a rare form of migraine headaches. It is even rarer to have a combination of isolated third cranial nerve palsy, all occurring in a patient of juvenile myoclonic epilepsy.

Although the association of migraine in patients with epilepsy and vice versa has been mentioned in medical literature frequently, it is rare to have the combination of isolated third cranial nerve palsy in ophthalmoplegic migraine in a patient of juvenile myoclonic epilepsy.

Previous reports of ophthalmoplegic migraine with cranial nerve palsies mentioned that among the cranial nerve, the third cranial nerve is the most common. The involvement of the recurrent sixth nerve and fourth nerves are well documented.

It is postulated that migraine headaches and epileptic seizures alike may be triggered by excessive neocortical cellular excitability and the association between the two is well documented. However, to the best of our knowledge, we have not found this combination of ophthalmoplegic migraine with isolated third cranial nerve palsy with juvenile myoclonic epilepsy in the same patient.

Figure 1 This collage of photographs of the patient's eye movements, at rest and in all vertical and horizontal gazes have been depicted. At rest, patient has left-sided ptosis; and at extreme right gaze, mild medial rectus restriction may be seen. Rest of the extraocular movements appeared normal.

Learning points

- ▶ Ophthalmoplegic migraine is a rare variant of migraine headaches.
- ▶ It may present with one or more cranial nerve involvement in combination or alone, namely third, fourth and sixth.
- ▶ The cranial neuropathies may be transient or recurrent, usually with complete recovery; however, residual deficits are known to occur.
- ▶ Epileptics have a higher chance of getting migraine headaches, and vice versa.
- ▶ Juvenile myoclonic epilepsy, a form of generalised epilepsy syndrome can present with ophthalmoplegic migraine with cranial nerve neuropathy.

Contributors AS was involved in the conception and design, or analysis and interpretation of the data. SK drafted the article or revised it critically for important intellectual content. AD gave the final approval of the version to be published.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed

REFERENCES

- 1 McMillan HJ, Keene DL, Jacob P, *et al.* Ophthalmoplegic migraine: inflammatory neuropathy with secondary migraine? *Can J Neurol Sci* 2007;34:349–55.
- 2 Bharucha DX, Campbell TB, Valencia I, *et al.* MRI findings in pediatric ophthalmoplegic migraine: a case report and literature review. *Pediatr Neurol* 2007;37:59–63.
- 3 Doran M, Larner AJ. MRI findings in ophthalmoplegic migraine: nosological implications. *J Neurol* 2004;251:100–1.
- 4 Shin DJ, Kim JH, Kang SS. Ophthalmoplegic migraine with reversible thalamic ischemia shown by brain SPECT. *Headache* 2002;42:132–5.
- 5 Neera C, Yogesh P, Vinod P, *et al.* Recurrent sixth nerve palsy in childhood ophthalmoplegic migraine: a case report. *Neurology Asia* 2013;18:95–8.
- 6 Alfradique I, Vasconcelos MM. Juvenile myoclonic epilepsy. *Arq Neuropsiquiatr* 2007;65:1266–71.
- 7 Hauser WA, Annegers JF, Rocca WA. Descriptive epidemiology of epilepsy: contributions of population-based studies from Rochester, Minnesota. *Mayo Clin Proc* 1996;71:576–86.
- 8 Delgado-Escueta AV, Enrile-Bacsal F. Juvenile myoclonic epilepsy of Janz. *Neurology* 1984;34:285–94.
- 9 Dhanuka AK, Jain BK, Daljit S, *et al.* Juvenile myoclonic epilepsy: a clinical and sleep EEG study. *Seizure* 2001;10:374–8.
- 10 Andermann E, Andermann FA. Migraine-epilepsy relationships: epidemiological and genetic aspects. In: Andermann FA, Lugaesi E, eds. *Migraine and epilepsy*. Boston, MA: Butterworths, 1987:281–91.
- 11 Leniger T, Diener HC. Migräne und Epilepsie—ein Zusammenhang? (Migraine and epilepsy—a relationship?). *Akt Neurol* 1999;26:116–20.
- 12 Ottman R, Lipton RB. Comorbidity of migraine and epilepsy. *Neurology* 1994;44:2105–10.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow