CASE REPORT

Acute confusional migraine: a variant not to be missed

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SUMMARY

A middle age woman who had frequent migraines was admitted with memory loss following severe unilateral headache, restlessness and confusion. Investigations including haematology, biochemistry, cerebrospinal fluid analysis serology and imaging were normal. As she had frequent migraines with a history of ophthalmoplegic migraine, acute confusional migraine was thought of as a possibility. The patient responded dramatically to intravenous sodium valproate. We report this case to emphasise that acute confusional migraine, a rare variant of migraine, is occasionally encountered in adults and prompt recognition will lead to appropriate management.

BACKGROUND

Acute confusional migraine (ACM) is a rare migraine variant. It is mainly seen in children with migraine. The incidence in paediatric age group is 0.45–7.8%. Adults are rarely affected. The presenting symptom of confusional state may be speech difficulties, decreased alertness, agitation and amnesia. Mild head trauma is thought to be the triggering factor in approximately 50% of the cases. ACM is essentially a diagnosis of exclusion as many life-threatening conditions like cerebrovascular accidents, intoxication, encephalitis and neoplasm also present as acute confusional state. Proper history, clinical examination and investigations are required to rule out other causes before making the diagnosis of ACM.

CASE PRESENTATION

A 29-year-old, right-handed woman was admitted with memory loss for 1 day. A day before, she had an acute onset of moderately severe, left-sided throbbing headache followed by restlessness, agitation and disorientation. She was given analgesics at home by her husband. The pain subsided to some extent and she slept. On waking up, she was neither able to recall the past events nor recognise her family members. She was aware of her personal identity and was able to carry out the activities of daily living including complex activities. She had frequent migraines with a history of status migrainosus 2 years previously and an episode of ophthalmoplegic migraine 1 year ago. There was no history of loss of consciousness, seizure, fever, weakness of any limb, swaying, drug overdose or poisoning. On examination, anterograde and retrograde amnesia were present. She also had total amnesia of the events which occurred during the attack. The rest of the neurological examination was normal.

A neuropsychological examination revealed immediate and recent memory impairment.

INVESTIGATIONS

Complete haemogram, blood sugar level, liver function test, renal function test, thyroid profile and serum vitamin B₁₂ level were within normal range. Cerebrospinal fluid (CSF) analysis showed normal glucose and protein level with no white cells. ELISA for IgM antibodies for herpes simplex virus was non-reactive. Antinuclear antibody was negative. EEG showed diffuse slow wave activity (figure 1). MRI of the brain was normal (figure 2).

DIFFERENTIAL DIAGNOSIS

The possible differential diagnoses of ACM are epileptic seizures, vascular disorders, encephalitis and intoxication. However, the clinical course with remission within hours and its rather typical presentation with migraine headache make it easier to distinguish. Biochemical investigations, CSF study, brain imaging and EEG have been carried out and excluded these possible causes.

TREATMENT

Intravenous sodium valproate was administered at a loading dose of 20 mg/kg.

OUTCOME AND FOLLOW-UP

On the next day, there was significant improvement, as she was able to recognise her family members and could recall the past events. On formal testing, there was no memory deficit. She was discharged on tablet divalproex sodium 500 mg/day as a prophylaxis. Repeat EEG was normal after 7 days.

DISCUSSION

We have described a case of amnesia following severe headache in a known migraineur. The amnesia responded to intravenous sodium valproate and the only deficit after 1 day of treatment was the inability to recollect the events during the headache. Acute confusional migraine is considered a rare migraine variant primarily seen in children and adolescents. Gascon and Barlow¹ for the first time used the term 'acute confusional migraine' (ACM) in their series of four paediatric patients. In the literature, only a few cases of ACM in adults have been published but confusional states have been described previously in adult patients with cerebral autosomal-dominant arteriopathy with subcortical infarcts and leucoencephalopathy (CADASIL).² Shahien *et al*³ reported a case of CADASIL

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Figure 1 EEG showing diffuse slow wave activity.

presented with migraine headache with aura and MRI of the brain was found to be suggestive of subcortical white matter hyperintensities.

Pietrini *et al*⁴ described this condition in four adult patients. The duration of attacks has been reported as short as 10 min to many hours and terminates with deep sleep. Upon waking up, the patients had normal mental status, but with a spotty

recollection of events during the period of confusion. Disorientation, memory problems, difficulty in recognising familiar people and speech disturbance are frequently observed.⁵

Our patient was presented with acute onset hemicranial headache and amnesia, which lasted for 24 h and relieved by pharmacological treatment. The pathophysiological mechanisms of ACM are still debatable. One of the hypotheses is that ACM

Figure 2 MRI of the brain (A) T-2 and (B) T-1 weighted with contrast image showing no significant abnormality.

is caused either by brainstem ischaemia or due to local cerebral oedema.^{6,7} Primary neuronal dysfunction, a result of spreading depression and secondary vascular changes, is considered as the cause by other authors.⁴ According to recent knowledge, ACM is found to be considered as a result of reversible generalised brain dysfunction mainly in the basal parts of the temporal lobes and the midline brain structures that are associated with 'cortical spreading depression' and with underlying vascular mechanisms. The underlying pathophysiological mechanism for migraine aura including complex auras is thought to be due to cortical spreading depression (CSD).⁸ Pietrini *et al*⁴ have proposed CSD as the pathophysiological mechanism in ACM. As the prevalence of ACM is probably as low as 0.4% in children, there is a scarcity of ictal investigations. However, most ictal abnormalities have been found in EEG studies in the form of slowing predominantly in frontal and occipital region.⁴ EEG was suggestive of diffuse slowing in our patient. Previous literature have demonstrated hypoperfusion in the left splenium of the corpus callosum in ictal SPECT⁹ in one patient and reversible narrowing of the left middle and posterior cerebral arteries in ictal MR angiography (MRA) in another patient.¹⁰ Attacks of ACM are usually self-limiting and is resolved by sleep; no specific treatment was used in most described cases.^{5,11} There have been few reports in the literature regarding the treatment of ACM. Avraham *et al*¹² reported a case of a young girl with a first episode of ACM that was responsive to valproic acid (VPA) during the acute confusional phase and intravenous VPA was suggested as an effective drug. In one of the preliminary studies carried out, Shahein *et al*¹³ concluded that intravenous sodium valproate was found to be safe and rapidly effective for intractable migraine attack. Intravenous sodium valproate has resolved the symptoms in our patient and no recurrence has been found on successive follow-up.

Learning points

- ▶ Acute confusional migraine (ACM) should be considered as a differential diagnosis, especially in a known migraineur presenting with acute onset amnesia or abnormal behaviour.
- ▶ ACM is a diagnosis of exclusion.
- ▶ Intravenous sodium valproate is the drug of choice for acute attack.

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