Recurrent Wernicke's Aphasia: Migraine and Not Stroke!

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We report the clinical findings of a 40-year-old woman with recurrent migraine presenting with Wernicke's aphasia in accordance with the results of a standardized battery for language assessment (Boston Aphasia Diagnostic Examination). The patient had no evidence of parenchymal or vascular lesions on MRI and showed delta and theta slowing over the left posterior temporal leads on the EEG. Although the acute onset of a fluent aphasia suggested stroke as a likely etiology, the recurrence of aphasia as the initial symptom of migraine was related to cortical spreading depression and not to stroke.

Key words: migraine, Wernicke, aphasia, EEG

INTRODUCTION

Migrainous aura usually comprises completely reversible visual and/or sensory and/or speech symptoms that have gradual development, progressive onset (5-20 minutes), and duration less than 60 minutes.[1] The underlying neural mechanism is considered to be cortical spreading depression (CSD), which produces symptoms like scintillating scotoma, sensory disturbances, aphasia, dysarthria, or hemiparesis. Here, we report the clinical findings of a patient who presented a Wernicke's aphasia as the initial symptom of migraine.

CASE DESCRIPTION

A 40-year-old French teacher suddenly developed a speech jargon characterized by incompre hensible words while giving a lecture, and was immediately brought to the emergency department of our hospital. Her past history was significant for recurrent headaches occurring 1-3 times every month since the adolescence. She believed there had been similar cases in her family. The pain used to be throbbing and hemicranial, usually without aura, occurred in association with nausea and photophobia, and was relieved by resting in a dark room or sleeping. Only once, 2 years previously, she had presented similar linguistic changes (verbal paraphasias and increased verbal fluency) during a migraine attack, which had lasted several hours.

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This time she presented with language difficulties, which were followed 30 minutes later by headache. Pain had progressive onset over the left cranial regions and was associated with nausea and photophobia. The patient was agitated, her language was fluent and characterized by verbal paraphasias (jargonaphasia) of which she seemed anosognosic, and presented severe difficulties in oral comprehension. This collection of symptoms was suggestive of Wernicke's aphasia, a diagnosis supported by the French adaptation of the Boston Aphasia Diagnostic Examination (BDAE-F)[2] (Table 1), which was carried out during the first hours after the onset of symptoms.

Aphasia persisted over 24 hours and disappeared gradually. The sudden onset of aphasia prompted us to search for an ischemic lesion. A brain CT (including perfusion scan and angio-CT sequences) performed one hour after the onset of symptoms did not show any signs of ischemia or vascular abnormalities. The following day, a brain MRI including DWI/perfusion sequences was performed and did not show any change. The CSF examination and routine blood tests were normal. Antiphospholipid and antinuclear antibodies were negative. The EEG, performed at the time when the patient produced paraphasias, showed irregular theta and delta slowing and a relative loss of alpha activity over the left posterior temporal and occipital regions (Fig. a). During drowsiness (Fig. b), sleep spindles showed an asymmetrical amplitude, left less than right.

Because of the absence of signs in favor of other neurological disorders, and because of the past history of migraine attacks with similar symptoms, the diagnosis of migraine with aura was finally selected in accord with the International Headache Society criteria.[1]

Conflict of Interest: None

Table 1.—Performance of the Patient on the Boston Aphasia Diagnostic Examination

Category	Subtests	Scores	Normal range
Auditory comprehension	Word discrimination	5†	N > 55 (72)
	Body part identification	2†	N > 15 (20)
	Command	1†	N > 10 (20)
	Logical reasoning	0†	N > 5 (12)
Fluency	Articulation rating	7	N > 6 (7)
	Phrase length	7	N > 5 (7)
	Verbal agility	n.t.	–
Automatic speech	Automatized sequences	n.t.	–
	Reciting	0†	N > 1 (2)
Repetition	Repetition of words	2†	N > 7 (10)
	Concrete sentences	0†	N > 5 (8)
	Abstract sentences	0†	N > 2 (8)
Reading aloud	Words	4†	N > 8 (10)
	Sentences	0†	N > 5 (10)
Naming	Responsive naming	0†	N > 22 (30)
	Confrontation naming	0†	N > 70 (105)
	Animal naming	2†	N > 7 (23)
	Body part naming	0†	N > 19 (30)
Paraphasias	Phonological and morphological	10†	N < 5
	Semantic	10†	N < 7
Written comprehension	Literal discrimination	2†	N > 7 (10)
	Auditory-written-word matching	4†	N > 6
	Spelled words	0†	N > 3 (8)
	Picture-written-word matching	3†	N > 7 (10)
	Text reading	2†	N > 5 (10)
Writing	Mechanics	3	N > 2 (3)
	Dictation	2†	N > 10 (15)
	Written denomination	0†	N > 5 (10)
	Graphical evocation	0†	N > 4 (10)
	Sentence dictation	0†	N > 5 (12)

The maximal possible score is reported in parentheses.
†Insufficient results.
n.t. = not testable.

DISCUSSION

Language-related symptoms before or during migraine have a prevalence of 5-22% among migrainous patients.[3,4] They are far less common, however, than visual symptoms (96.5% prevalence).[5] Aphasic changes occur more frequently (47%), are more severe, and last longer when associated with hemiplegic migraine.[6] The language or speech symptoms occurring during a migraine attack with aura appear to be hard to categorize,[1] but include dysarthria,[1] stuttering,[7] nonfluent or Broca's aphasia,[8,9] and anomia.[4]

To the best of our knowledge, there are no previous reports of patients presenting Wernicke's aphasia as the migrainous aura. In our patient, the diagnosis of Wernicke's aphasia was supported by the use of a standardized battery for language assessment (BDAE-F). The assessment of aphasia and speech disturbances occurring with migraine is rarely performed with standardized tests, which explains the paucity of quantitative and qualitative data in the literature about these disturbances.

Wernicke's aphasia is usually the clinical feature of ischemic stroke involving the auditory association cortex. For the patient we described, however, there were no signs of ischemia on the neuroimaging undertaken (including perfusion MRI).

Radiological changes (cortical edema in the majority of cases) associated with aura have been reported only in patients with persistent or prolonged aura (more than 4 hours)[10,11] and in a minority of patients with hemiplegic migraine.[12,13] Usually, these anomalies are diffuse and do not allow a correlation between clinical symptoms and defined arterial territories.

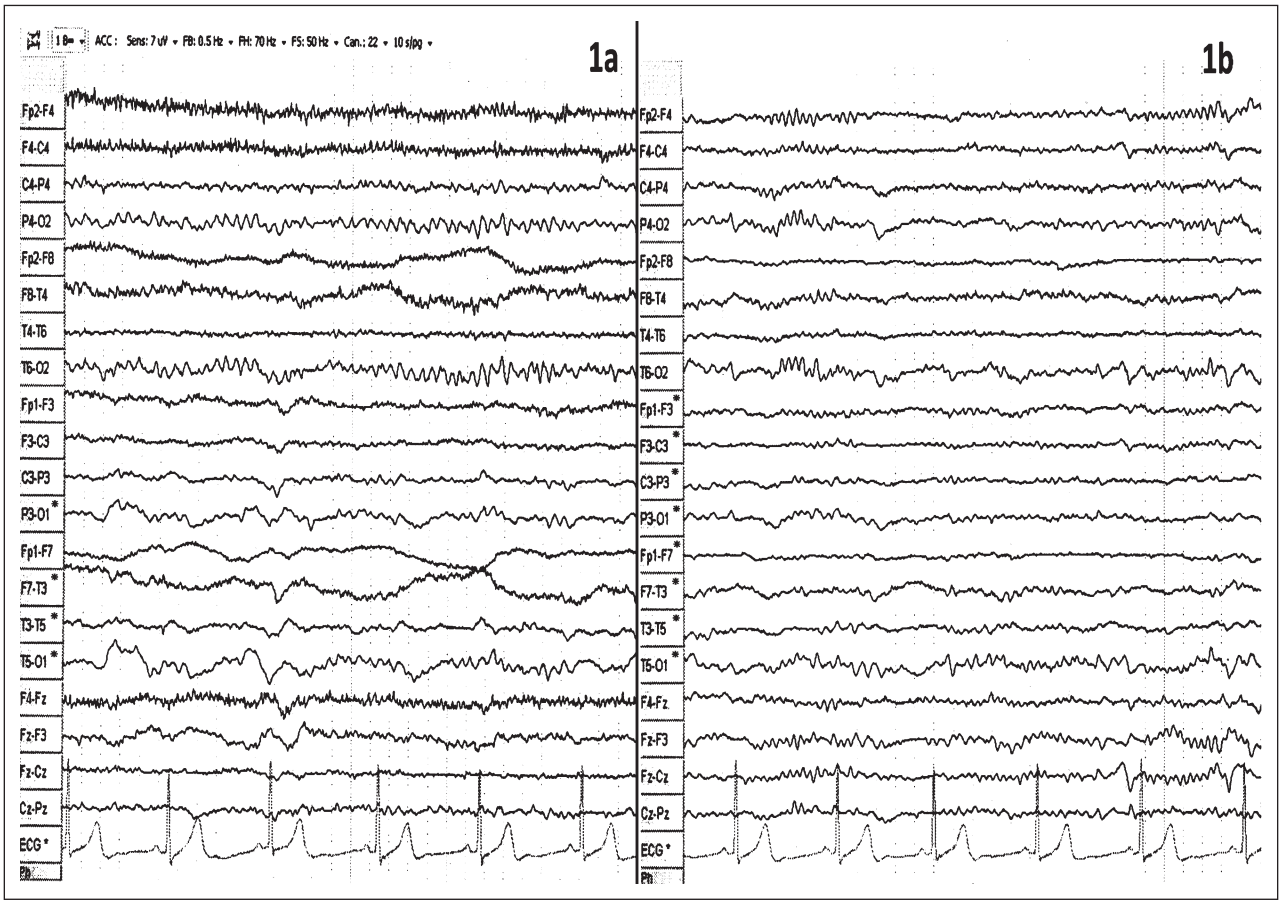

Fig.—EEG examination: bipolar, longitudinal montage (right over left, midline at the bottom). Paper speed 30 mm/sec, HFF: 70 Hz, LFF: 0.5 Hz, notch filter: 50 Hz, sensitivity: 70 μV/cm. (a) Posterior 9 Hz alpha on the right, and an irregular theta-delta slowing over the left hemisphere mostly involving the temporal leads (black dots). (b) Asymmetric distribution of the sleep spindles with a reduced amplitude on the left hemisphere (black dots).

In our patient, an EEG provided the evidence of cortical dysfunction over the left temporal regions.

The symptoms of the migrainous aura are generally related to the phenomenon of CSD that consists of depolarization of neuronal and glial cells (propagating in the cortex at the rate of 2-3 mm per minute), inducing a period of depression of the neuronal activity. Animal studies suggest that CSD can lead to hypoxia, neuronal swelling, increased extracellular potassium, and loss of dendritic spines (see Eikermann-Haerter and Moskowitz[14] for a review).

These metabolic changes can slow EEG activity.[15]

In conclusion, the clinical picture we reported indicates that it is possible to perform linguistic studies in migrainous patients who present with a language disorder in order to better localize the site of brain dysfunction. The factors that predispose certain specific brain regions to be implicated more than others during the migraine attacks remain poorly defined and constitute an interesting field for future research.

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REFERENCES

1. The International Headache Society Classification Subcommittee. The international classification of headache disorders: 2nd edition. *Cephalalgia*. 2004; 24(Suppl. 1):9-160.
2. Mazaux J, Orgogozo I. *BDAE-F. Boston Diagnostic Aphasia Examination*. Issy les Moulineaux: Editions Scientifiques et Psychologiques; 1981.
3. Bana DS, Graham JR. Observations on prodromes of classic migraine in a headache clinic population. *Headache*. 1986;26:216-219.
4. Vincent MB, Hadjikhani N. Migraine aura and related phenomena: Beyond scotomata and scintillations. *Cephalalgia*. 2007;27:1368-1377.

5. Olesen J, Friberg L, Olsen TS, et al. Timing and topography of cerebral blood flow, aura, and headache during migraine attacks. *Ann Neurol*. 1990; 28:791-798.
6. Bradshaw P, Parsons M. Hemiplegic migraine, a clinical study. *Q J Med*. 1965;34:65-85.
7. Perino M, Famularo G, Tarroni P. Acquired transient stuttering during a migraine attack. *Headache*. 2000;40:170-172.
8. Kirchmann M. Migraine with aura: New understanding from clinical epidemiologic studies. *Curr Opin Neurol*. 2006;19:286-293.
9. Linn J, Freilinger T, Morhard D, Bruckmann H, Straube A. Aphasic migraineous aura with left parietal hypoperfusion: A case report. *Cephalalgia*. 2007;27:850-853.
10. Smith M, Cros D, Sheen V. Hyperperfusion with vasogenic leakage by fMRI in migraine with prolonged aura. *Neurology*. 2002;58:1308-1310.
11. Relja G, Granato A, Ukmar M, Ferretti G, Antonello RM, Zorzon M. Persistent aura without infarction: Description of the first case studied with both brain SPECT and perfusion MRI. *Cephalalgia*. 2005;25:56-59.
12. Dreier JP, Jurkat-Rott K, Petzold GC, et al. Opening of the blood-brain barrier preceding cortical edema in a severe attack of FHM type II. *Neurology*. 2005;64:2145-2147.
13. Cha YH, Millett D, Kane M, Jen J, Baloh R. Adult-onset hemiplegic migraine with cortical enhancement and oedema. *Cephalalgia*. 2007;27:1166-1170.
14. Eikermann-Haerter K, Moskowitz MA. Animal models of migraine headache and aura. *Curr Opin Neurol*. 2008;21: 294-300.
15. Takano T, Tian GF, Peng W, et al. Cortical spreading depression causes and coincides with tissue hypoxia. *Nat Neurosci*. 2007;10:754-762.

Nasal Sumatriptan as Adjunctive Therapy for idiopathic Trigeminal Neuralgia: Report of Three Cases

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We examine the effect of nasal sumatriptan as an adjunctive therapy in 3 patients with idiopathic trigeminal neuralgia refractory to carbamazepine (CBZ) and found that this therapy might be suitable for patients for whom the CBZ dose cannot be increased, who are under poor pain control, and who are not candidates for nerve blocks or surgery.

Key words: nasal sumatriptan, idiopathic trigeminal neuralgia, adjunctive therapy

INTRODUCTION

Idiopathic trigeminal neuralgia (ITN) is a type of facial pain characterized by sudden attacks or severe, relatively short-lasting bouts of pain that are often described as electric-like.[1] The pain is commonly evoked by trivial stimuli such as touch, chewing, and talking. The therapies for ITN are divided into pharmacotherapy, using antiepileptic drugs such as carbamazepine (CBZ), phenytoin, or gabapentin, and nerve blocks and microvascular decompression.[2] CBZ is the only drug that has been subjected to several randomized controlled trials in large patient populations,[3,4] although some patients discontinued it because of serious adverse effects.

A recent study that was conducted in 24 patients with ITN refractory to previous therapies demonstrated that subcutaneous injection of sumatriptan, a 5-$HT_{1B/1D}$ receptor agonist, produced prompt and continuous analgesia.[5] This finding suggests the possibility that subcutaneous sumatriptan might be an effective treatment for ITN, since it prevents vasodilation and inflammation of the irritated trigeminal root.[6,7] The treatment is invasive and very painful, however, and requires self-injection. To overcome these disadvantages of subcutaneous sumatriptan, we investigated the effect of nasal sumatriptan as adjunctive therapy for patients with ITN refractory to CBZ.

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