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Nonconvulsive status epilepticus presenting as a subacute progressive aphasia

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We report a 62-year-old man with non-convulsive status epilepticus (NCSE) presenting as a progressive aphasia that developed insidiously over 5 weeks. On video-EEG monitoring, aggravation of the aphasia coincided with occurrence of seizure activities arising from the left fronto-temporal area. Brain MRI was noncontributory but a fluorodeoxyglucose-PET scan revealed a hypometabolism in the left anterior temporal area. Following anticonvulsant treatment, aphasia recovered gradually over several weeks despite prompt resolution of epileptic discharges on EEG. Our patient's findings, gradual onset of isolated aphasia with gradual resolution after initiation of treatment, may differ from previously reported cases with aphasic status epilepticus because their aphasia showed abrupt onset and rapid resolution with anticonvulsant medication.

Key words: non-convulsive status epilepticus; aphasia; positron emission tomography.

INTRODUCTION

Aphasia or speech disturbance is a common ictal or post-ictal symptom of seizures[1]. However, aphasia as the sole manifestation of seizure is rarely reported[1–10] because it is frequently associated with other symptoms such as altered consciousness or convulsive movements. Rosenbaum *et al.*[1] defined ictal aphasia using the following criteria. First, the patient must be capable of speaking during the ictus. Second, the speech produced must be aphasic (i.e. dysfluent, dysnomic, or paraphasic). Third, consciousness is not impaired. Lastly, ictal EEG recording must demonstrate abnormal discharges that correlate with clinical events.

Prolongation of ictal aphasia is referred to as aphasic status epilepticus. In previously reported cases, aphasic status epilepticus was of relatively abrupt onset and resolved rapidly with anticonvulsant medication[1–3, 6–10]. Unlike these patients, our patient with nonconvulsive status epilepticus (NCSE) showed gradual evolution of isolated aphasia over several weeks and protracted recovery of language over a month after initiation of treatment.

CASE REPORT

A 62-year-old right-handed man with a formal education of 16 years presented with a speech disturbance that progressed over 5 weeks. He had been relatively healthy, working as an assistant in a lawyer's office. Five weeks prior to admission, he developed occasional paraphasic errors and clumsy handwriting. Several days later his speech became slurred. However, he was able to maintain his job until 15 days before admission, when he had moderate difficulty in conversation due to slurred speech. Thereafter, his speech disturbance worsened relentlessly. According to his wife, he frequently burst into tears and complained of headaches. His past medical history was remarkable for diabetes mellitus and liver cirrhosis associated with viral hepatitis. He had no previous history of hypertension, stroke, seizure disorder or head trauma. He was a social drinker and non-smoker.

On admission, physical examinations including vital signs were unrevealing. On neurological examination he was alert and attentive. However, his spontaneous speech was slurred, laborious and halting, consisting of one- or two-word sentences. Auditory

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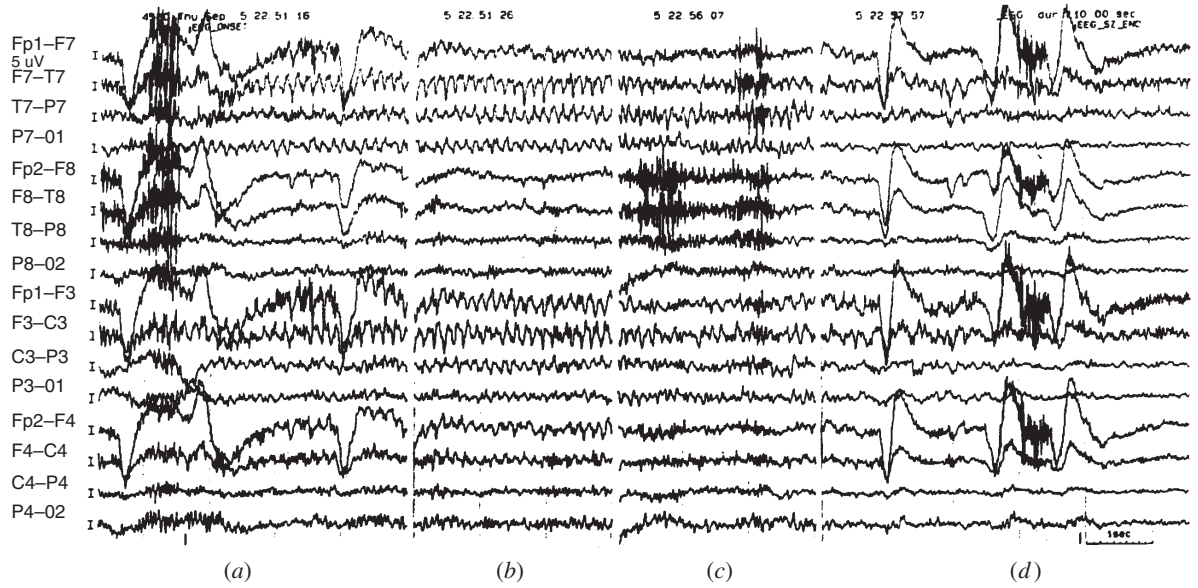

Fig. 1: Ictal EEG shows 5–7 Hz rhythmic waves arising mainly from the left temporal and frontal areas (*a*), building up in the same regions (*b*), and losing its rhythmicity as the seizure terminates (*c*, *d*). The EEG seizure was only accompanied by aggravation of aphasia. The filter settings were high frequency of 70 Hz and low frequency of 1 Hz.

comprehension was also severely impaired as were repetition, naming and reading. Writing was also markedly impaired, showing unintelligible scribbling movements. He appeared to be aware of his speech disturbance and seemed depressed. Cranial nerves, sensory and motor examinations were normal. Tendon reflexes were within normal limits and symmetric with flexor plantar response. On the day of admission, he had a seizure starting with right facial twitching and focal clonic movement in his right arm, evolving to right head version and rightward gaze deviation, followed shortly after by a generalized tonic–clonic convulsion lasting a few minutes.

Routine blood work-ups were remarkable for mild thrombocytopenia (70 000 u L^{-1}), elevated AST and ALT (59 and 49 u L^{-1}), prolonged prothrombin time (INR, 1.28) and a high fasting serum glucose of 339 mg dL^{-1} with HbA1C of 14.7. Other blood tests including serum ammonia and electrolytes were all normal. Lumbar puncture yielded acelluar fluid with normal glucose and protein. PCRs for herpes virus and tuberculosis were negative.

Routine scalp EEG on the day of admission showed intermittent slowing and an electroencephalographic seizure originating from the left hemisphere. On the same day, video-scalp EEG monitoring was performed for 12 h. During this study, he had three episodes of aggravation of language disturbance, characterized by global aphasia without altered consciousness with no postictal confusion. The episodes lasted several minutes and coincided with seizure activities originating from the left fronto-temporal area (Fig. 1). During times when there was no electroencephalographic seizure activity, he was able to produce one- or two-word sentences and comprehend simple commands (e.g. close your eyes). Brain MRI showed only small foci of abnormal signal interpreted as lacunes in the pons and left thalamus. Additionally, T2-weighted images showed a subtle region of reduced signal intensity in the white matter of the left anterior temporal region (Fig. 2).

After completion of the video-EEG monitoring, diphenylhydantoin was loaded orally (300 mg three times every 2 h on the 2nd hospital day) and its serum levels were 13.9, 21.0 and 12.7 μg ml^{-1} on the 5th, 8th and 15th hospital day, respectively. After initiation of treatment, his language disturbance improved gradually over several weeks (see below). However, routine scalp EEG performed on the third hospital day revealed neither seizure activity nor epileptiform discharges aside from intermittent slow waves in the left temporal area. The slow waves disappeared on a repeated EEG performed on the 7th hospital day. FDG–PET performed on the 13th hospital day showed glucose hypometabolism in left anterior temporal lobe.

One month after discharge (50 days after initiating treatment) repeated routine scalp EEG with sphenoidal electrodes was normal and his speech was nearly normalized. By 4 months after discharge, his language had completely recovered. Repeated brain MRI was unchanged except that the low signal in the white matter of the anterior temporal lobe had resolved. A repeated FDG–PET scan performed at that time showed aggravation of the left anterior temporal hypometabolism (Fig. 2).

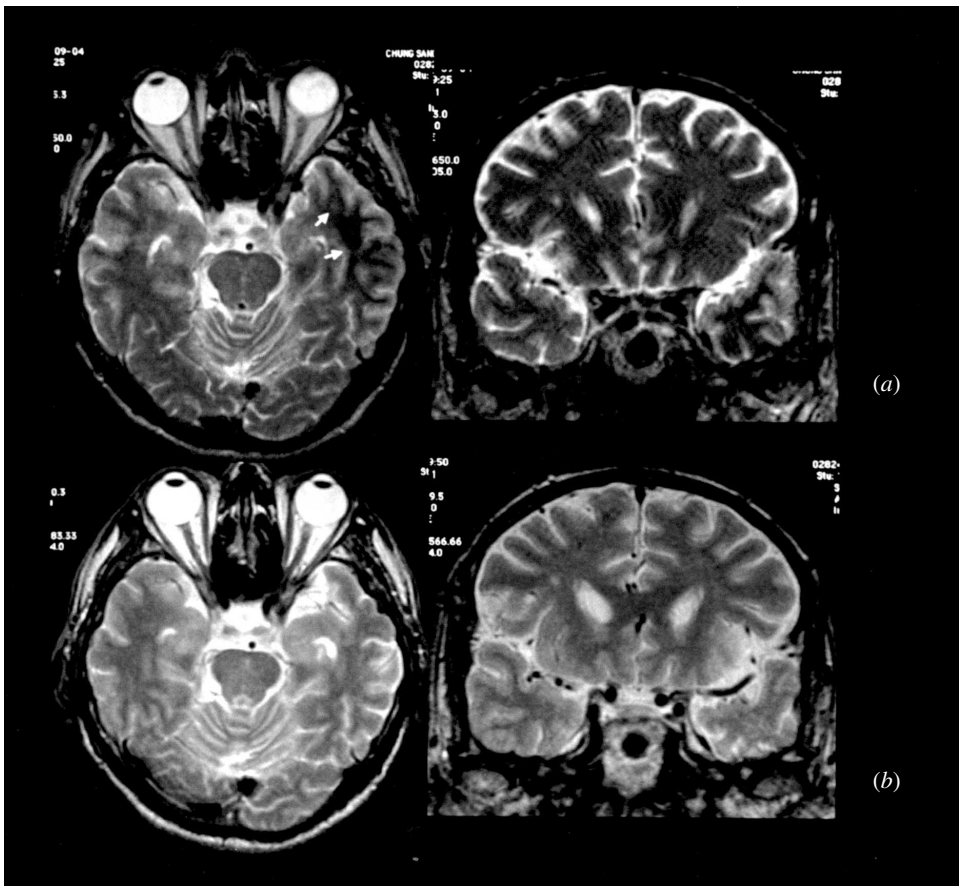

Fig. 2: Brain MRI was performed during admission (*a*) and 4 months later (*b*). Initial T2 MRI shows no structural abnormalities except for a low signal area in the left temporal lobe (arrow) which disappeared on the follow-up MRI.

Table 1: Results of Western aphasia battery and BNT after treatment with antiepileptic drug.

Interval after treatment	Spontaneous speech		Comprehension (10)	Repetition (10)	Naming (10)	AQ[a] (100)	BNT (60)
	Information (10)[b]	Fluency (10)					
3 days	0	4	0.65	0	0	9.3	NA[c]
24 days	9	8	9.0	9.4	9.0	88.8	19/60
3 months	10	10	10.0	10.0	10.0	100.0	51/60

[a] AQ: aphasia quotient, [b] Scores in parentheses are maximum scores, [c] NA: not available.

The patient's language function was evaluated with the Korean version of Western Aphasia Battery (K-WAB)[11] and the Korean version of the Boston Naming Test (K-BNT)[12] (Table 1). On the fourth hospital day (3 days after treatment) spontaneous speech was limited to automatic speech phrases (e.g. 'I don't know'). Other language functions, including reading and writing, were unchanged. On the seventh hospital day, he could speak one phrase level and repeat up to three syllable words. At discharge on the 25th hospital day, he still showed hesitancy with word-finding difficulty during conversational speech, although he scored an 8 out of 10 possible fluency rating. He was able to comprehend and repeat quite well, but the result of K-BNT was still below the first percentile (19/60). Three months later, his performance on the K-WAB was flawless and his performance on the K-BNT was also over the 99th percentile (51/60). The patient returned to his job but still noted difficulty expressing himself when speaking rapidly.

DISCUSSION

Our patient presented with nonfluent aphasia that developed insidiously and progressed over several weeks. Seizure had hardly been considered in the

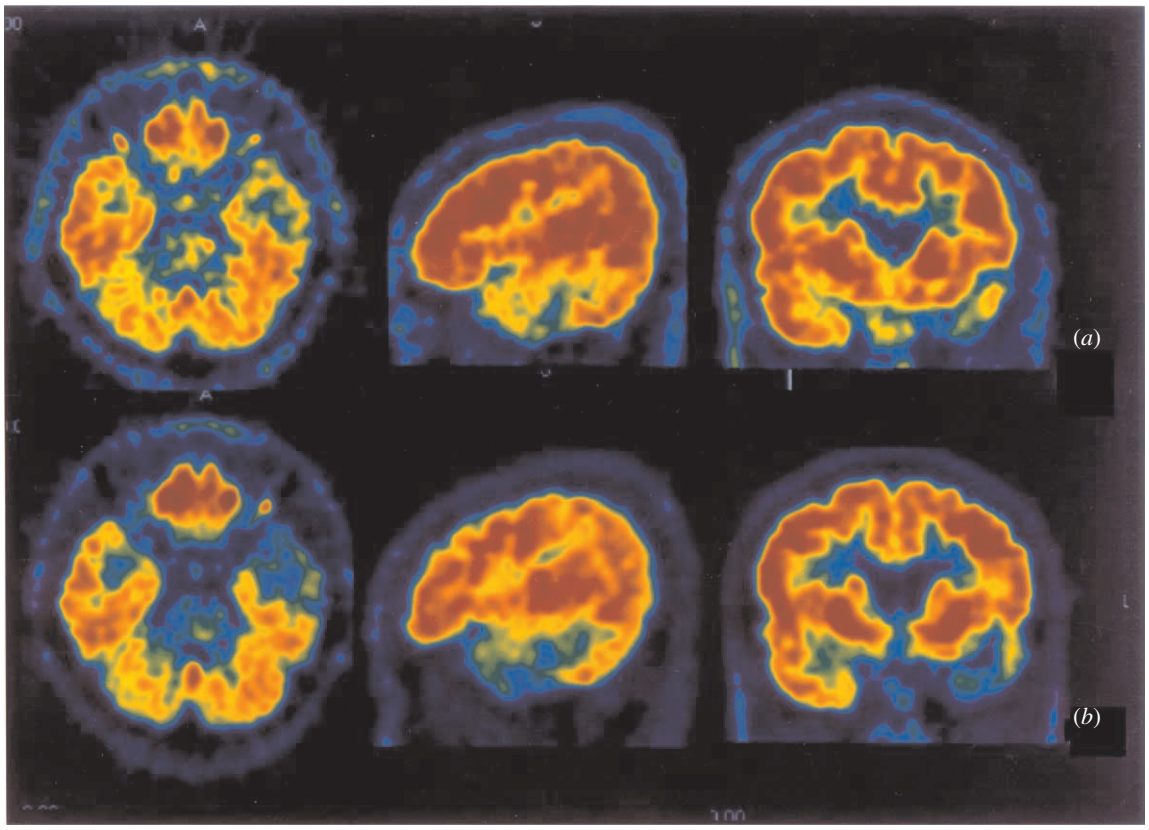

Fig. 3: FDG–PET was performed during admission (*a*) and 4 months later (*b*). FDG–PET demonstrates glucose hypometabolism in the left anterior temporal lobe which aggravated on follow-up PET despite clinical improvement.

differential diagnosis until the patient developed the focal convulsions with secondary generalization on the day of admission. The patient's aphasia fulfilled the criteria of ictal aphasia proposed by Rosenbaum *et al.*[1] While the patient was aphasic, he demonstrated no convulsive movements except for the single focal motor seizure. Video-scalp EEG monitoring confirmed that aggravation of aphasia coincided with the occurrences of epileptic discharges arising mainly from the left fronto-temporal area. Therefore the persistent aphasic phenomenon observed in our patient may represent a form of NCSE.

Table 2 summarizes aphasia cases from NCSE that have been reported so far[1–3, 6–10]. In previously described cases, the aphasia developed suddenly and usually resolved promptly when seizures were controlled. Unlike these cases, however, our patient took the clinical course of subacute progression and then gradual recovery after treatment. Insidious onset and progression of the aphasia may represent ictal or postictal deficits from unrecognized partial seizures occurring at an increasing frequency before culminating in NCSE. Gradual recovery in our patient might be due to either incomplete seizure control or an underlying structural lesion rather than seizure *per se*. However, EEG abnormalities were promptly normalized following administration of the antiepileptic drug and the MRI demonstrated no significant structural lesions other than a subtle low signal in the white matter of the anterior temporal lobe.

The patient's initial language impairment was classified as global aphasia. Anatomical lesions causing global aphasia are usually extensive involving both Broca's and Wernicke's areas. Our patient, however, had a localized functional deficit zone in the left anterior temporal lobe identified by PET scanning. One parsimonious account for this discrepancy might be the anatomical proximity of our patient's lesion to the basal temporal language area (BTLA). Lüders *et al.* discovered the BTLA during electrical stimulation of the cortex in epileptic patients[13]. Other studies have confirmed the existence of the BTLA as a language center[14, 15]. The BTLA is located in the occipitotemporal gyrus, and partly in the inferior temporal and parahippocampal gyri, extending up to ~6–7.5 cm posteriorly from the tip of the temporal lobe[14, 15]. Language disturbance associated with electrical stimulation of the BTLA ranges from isolated anomia to global aphasia[14]. Abou-Khalil *et al.* also reported a patient with global aphasia resulting from a seizure focus in the left basal temporal region[16]. An alternative account for the

Table 2: Summary of previously reported cases with aphasic status epilepticus.

Author	Type of aphasia	Etiology	Previous seizure	Onset/course	Duration of aphasia before treatment	Time from treatment to recovery of aphasia	Other symptoms
Racy *et al.*[7], 1980							
Case 1	Global	Unknown	No	Sudden/wax and wane	1 day	3 days	No
Case 2	Wernicke	Lt. parietal glioblastoma	No	Sudden/persistent	3 days	2 days	Convulsive seizure
Dinner *et al.*[8], 1981	Global	Unknown	No	Sudden/wax and wane	10 days	12 days	Memory loss Convulsive seizure
Rosenbaum *et al.*[1], 1986	Global	Lt. parietotemporal infarction	No	NA[a]/wax and wane	1 week	4 days	Rt. hemiparesis, Rt. side clonic seizure
Primavera *et al.*[9], 1988	Global	Lt. temporal old ICH	No	NA[a]/wax and wane	15 days	10 days	No
Wells *et al.*[6], 1992	Global	Lt. temporal glioblastoma	No	Sudden/persistent	1 day	?Rapid response	Rt. pronator drift
Kirshner *et al.*[2], 1995	Wernicke	Unknown	No	Sudden/persistent	4 days	Several days	No
Primavera *et al.*[3] 1996	Global	Multiple sclerosis	Yes	Sudden/persistent	Several hours	5 days	Rt. side clonic seizure
Grimes & Guberman[10], 1997	Global	R/O Lt. temporoparietal ischemic stroke	No	Sudden/persistent	3 days	?Rapid response	Rt. gaze preponderance
Our case	Global	Unknown	No	Gradual/progressive	30 days	Several weeks	No

[a]NA: not available.

global aphasia might be that the patient had temporal lobe seizures resulting in postictal dysphasia due to the spread of the ictal and postictal dysfunction into the language centers in the left hemisphere.

Despite complete resolution of aphasia in our patient, follow up PET paradoxically showed aggravation of hypometabolism in the left anterior temporal area while repeat MRI was largely unchanged. Although the histological identity of this lesion was not confirmed, it is less likely to be brain tumor or infectious etiology. Rather, it may be attributable to brain injury associated with status epilepticus as has been suggested in earlier investigations[17]. Alternatively, there might have been a subclinical vascular insult either preceding or coinciding with onset of the seizures and language disturbance. The MRI changes associated with status epilepticus include increased signal intensity on T2- or diffusion-weighted images in the affected cortex or underlying white matter[18]. Hence, the nature of the focus of low signal intensity in our patient's temporal subcortical white matter remains unclear.

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