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Case Report

De novo aphasic status epilepticus: Finally making the diagnosis by long-term EEG

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ABSTRACT

Aphasic status epilepticus (SE) is a rare manifestation of non-convulsive SE (NCSE) and may occasionally be under-recognized. We report a 69-year-old male patient with a pre-existing left parietal oligodendroglioma WHO III after two resections and radio-chemotherapy. The patient was left with some word finding difficulties but had no history of overt seizures. He developed aphasic NCSE, which was only detected by long-term electroencephalography (EEG) monitoring. The 24-hour EEG revealed paroxysmal rhythmic theta-delta activity in left posterior regions that propagated to left temporo-parietal areas. Rhythmic activity appeared every 15–30 min and lasted for 10–110 s. Aphasia was continuously present with superimposed short-lasting clinical deteriorations during the day. Magnetic resonance imaging showed peri-ictal edema on diffusion-weighted images in the insula and fronto-parietal cortex, which supported the diagnosis of SE. NCSE persisted for seven months. The patient recovered upon addition of intravenous phenytoin. One should not only consider aphasic SE when language impairment is episo-dic, but also when there are prolonged manifestations, especially when the typical differential diagnoses have been excluded. Intravenous therapy may be required to terminate NCSE. With this report, we would like to draw attention to aphasic SE as a rare phenomenon that may be difficult to diagnose and delay management in clinical practice.

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Introduction

Nonconvulsive status epilepticus (NCSE) is a heterogeneous disorder of varied etiologies and several subtypes. It is a common, yet still under-recognized condition, especially in the elderly [1]. The global incidence of both convulsive and nonconvulsive status epilepticus (SE) is 26.2 per 100,000 in the elderly population versus 5.2 per 100,000 in the younger population [1]. Diagnosis is important for adequate treatment, and delay in diagnosis and treatment may be associated with increased mortality [1]. NCSE is usually not

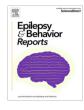
Abbreviations: ASM, antiseizure medication; CTP, CT-perfusion; DWI, diffusionweighted imaging; EEG, electroencephalography; FDG-PET, fluorodeoxyglucose positron emission tomography; FLAIR, Fluid-attenuated inversion recovery; MRI, Magnetic resonance imaging; LPD, lateralized periodic discharges; NCSE, nonconvulsive SE; SE, status epilepticus; PCV, Procarbazin/Lomustin/Vincristin. * Corresponding author.

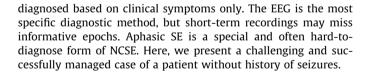
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Case report

This right-handed patient was diagnosed with a left parietal oligodendroglioma WHO III°, which was resected when he was 48 years old. He received radio-chemotherapy (two cycles of Procarbazine/Lomustin/Vincristin, PCV), which was subsequently discontinued due to hematological complications. The patient relapsed at the age of 60 in the left temporal lobe, and the tumor was re-resected, followed by a brief chemotherapy course with two cycles of PCV, which was also terminated prematurely due to complications. According to the treating neurosurgeon, the only available source for the patient's post-interventional performance, he was left with slight word finding difficulties and a mild impair-



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ment of his short-term memory. According to the patient's relatives, these deficits did not increase in the following years and the patient did not experience seizures.

At the age of 69, the patient suddenly became aphasic. According to the relatives, he complained of a headache a few hours before the onset of aphasia, which started when the patient was awake; the relatives could not recollect more precise pieces of information seven months after this event, and the local hospital notes were also not informative. In the patient's local hospital, ischemia, tumor recurrence, or encephalitis were excluded with MRI and cerebrospinal fluid analysis. Magnetic resonance imaging (MRI) revealed no new structural lesions (Fig. 1A). On the standard EEG over 15 minutes, only left temporo-occipital slowing was described. In the absence of a better explanation and a degree of suspicion for seizures, the patient was started on antiseizure medication (ASM) with levetiracetam, which was extended on an outpatient basis with lacosamide and valproic acid. Since the language problems persisted, he was admitted to our center seven months after onset of the aphasia. The clinical examination revealed fluctuating mild-to-moderate right-sided hemiparesis and global aphasia. There was some degree of swallowing difficulties. The patient was only able to give his name and place of residence and follow simple verbal instructions. The patient was only able to give his name and place of residence and follow simple verbal instructions, such as arm lifting and walking when he was asked to. He was not able to build a sentence or read text. Simple questions could be answered with yes or no. He could only name a few objects, such as "watch" or "table". He could not follow body commands, for example touching the ear with the right hand, were not possible to perform. He was awake and could fix gaze. There were no other subtle clinical phenomena like periorbital or perioral myoclonia. During the day, there was a recurring deterioration in language production. For several minutes at a time, the patient was unable to say his name or name his place of residence. Upon admission, the patient received treatment with three antiseizure medications including lacosamide 300 mg/day (trough blood level, 7.4 µg/ml), brivaracetam 200 mg/day (trough blood level, 2.07 µg/ml) and valproic acid 1500 mg/day (trough blood level, 64 µg/ml) without obvious side effects.

The long-term EEG without ASM reduction and without online supervision showed a continuous left parieto-occipital theta slowing with rare intermingled sharp waves. Within 24 h, rhythmic theta-delta activity occurred every 15–30 min with a maximum in the left posterior temporal lobe (T5) and lasted for 10–110 s. It propagated to the left temporo-parietal electrodes (T3 and P3), as displayed in Fig. 2.

A discontinuous aphasic SE was diagnosed. Benzodiazepines (one single dose of 2 mg intravenous lorazepam and oral clobazam 20 mg/day) were initially used. No clinical improvement was noted, despite rarer and shorter ictal EEG patterns. Brain MRI showed the partially resected tumor with normal diffusion-weighted imaging (DWI, Fig. 1E). In retrospect, DWI from onset of the aphasia (Fig. 1B) and 2.5 months later (Fig. 1D) revealed a signal increase in the insula and frontoparietal cortex. These changes and their courses had been previously overlooked and were now interpreted as ictal phenomena.

Seventeen days after admission, vigilance decreased during benzodiazepine treatment. The patient developed an aspiration pneumonia that was deemed to be a complication of altered vigilance under treatment with benzodiazepines. His condition required intensive care treatment for three days. In the intensive care unit, oral anticonvulsant therapy was switched to intravenous delivery, and phenytoin was carefully added (6.8 mg/kg body weight). This resulted in regression of the aphasia. ASM therapy was subsequently simplified to oral phenytoin 300 mg/day (16.5 μ g/ml) and brivaracetam 200 mg/day (0.92 μ g/ml), both in

two divided doses. He was discharged two months after admission and was back to his performance level prior to the aphasia (i.e., experienced mild word-finding problems, a mild right-sided hemiparesis, and no swallowing problems). Upon follow-up seven months after discharge, he was stable on the same medication. Long-term EEG continued to reveal subclinical seizure patterns (one/hour), but MRI returned to baseline (Fig. 1F, G).

The patient gave written informed consent for publication of this report and associated figures.

Discussion

In this case, the diagnosis of aphasic NCSE was challenging and defied early detection. Only long-term EEG permitted the diagnosis and finally led to intravenous phenytoin treatment, which restulted in termination of the status. Such a late but successful treatment appears noteworthy due to the duration of SE. In retrospect, MRI provided support by demonstrating hyperintense DWI abnormalities that normalized after beginning more intense treatment.

Making the diagnosis of aphasic SE

Diagnostic criteria for aphasic seizures were suggested by Rosenbaum et al. [2] and modified by Grimes and Gubermann [3], as displayed in Table 1. Our case satisfies these criteria.

The Salzburg criteria for NCSE [4], in general, are also fulfilled in our case since there were repetitive, rhythmic theta-delta activity with uniform morphology and duration with typical spatiotemporal evolution and clinical improvement after intravenous ASM (in our case, phenytoin) with EEG improvement (seizure patterns occurred less frequently) [5]. Rhythmic EEG activity was continuously present for at least 10 s (in our case, up to 110 s), which increased the specificity. Despite the previous trephination, the ictal patterns were not strongly expressed. This raises the question of a more deep-seated ictal focus, which might fit with the insular involvement on MRI. Anterior insular dysfunction contributes to aphasia [6].

The present case supports the notion that standard EEG recordings may not be sufficient for making the diagnosis in discontinuous status epilepticus. In one study, status epilepticus was determined in five of nine (56%) situations with the standard EEG (30 min), while the other four cases could only be diagnosed using long-term EEG [7].

A similar phenomenon has been described in another study [8]. The aphasic status in two patients were difficult to diagnose, as the initial routine EEG gave inconclusive results. Only repeat EEGs revealed epileptiform activity in both cases.

In a small group of patients (N = 5) with normal EEG and brain MRI, in which aphasic SE was suspected, fluorodeoxyglucose positron emission tomography (FDG–PET) confirmed the diagnosis by revealing hypermetabolism in language areas [9]. Two groups [9,10] suggested, in the case of clinical suspicion of aphasic SE in which EEG failed to show clear ictal activity and conventional MRI was normal, other methods, such as FDG–PET or, in acute situations, CT-perfusion (CTP). The findings of hypermetabolism in the epileptogenic zone on FDG–PET or hyperperfusion on CTP in the dominant hemisphere can lead to the diagnosis.

An additional tool for diagnosing NCSE due to a structural process may be DWI [11]. Our case, in retrospect, had increased DWI signal for at least 2.5 months.

Patients with Focal SE and peri-ictal DWI restriction often present with lateralized periodic discharges (LPDs) on EEG [12]. These LPDs rarely occur in isolation. Often, there is an intermittent transition into seizure patterns [13]. One case with aphasic SE with LPDs over the left frontocentral region in the continuous EEG had

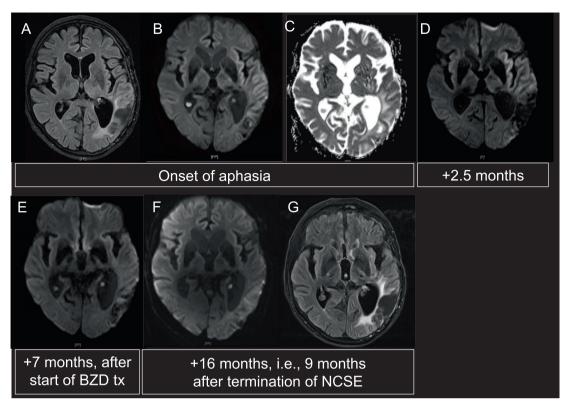


Fig. 1. Serial brain magnetic resonance images (MRI), axial sections. (A) Fluid-attenuated inversion recovery (FLAIR) at onset of the aphasia (B) Diffusion-weighted imaging (DWI) sequences from the same study as (A), showing hyperintense signal in the insula and in the frontoparietal cortex, (C) accompanied by cortical hypointensity on maps of the apparent diffusion coefficient (ADC). There is hyperintense "T2 shine through" in the neighbouring white matter. In summary, these images support clinical features of status epilepticus with a focal cortical diffusion restriction. (D) DWI 2.5 months later, still with similar hyperintensity. (E) After start of benzodiazepine treatment, the DWI abnormalities disappeared. (F) DWI image seven months after discharge was normal. (F) FLAIR was unchanged compared to (A). Abbreviations: BZD, benzodiazepine; NCSE, non-convulsive status epilepticus; tx, therapy.

a left frontal structural lesion after a meningioma operation and no history of seizures. Nevertheless, she was placed on ASM with phenytoin. Phenytoin serum level was initially low, and after increasing the dose, aphasia resolved completely [14]. In one retrospective study, 10% of the aphasic SE EEGs showed LPD [10]. The diagnosis of aphasic SE becomes challenging in these cases, as these patterns are not included in the EEG criteria for NCSE [5].

Onset and course of aphasic SE

In one study, the onset was acute in four of seven patients. In the remaining three, it was more gradual and fluctuating [8]. The duration of aphasia ranged from one hour to 20 days on admission, and the duration of recovery after SE treatment lasted from minutes and hours to days and weeks [10].

In this patient, we could not document a clear correspondence of presence of absence of a status pattern on EEG, especially since there was no online supervision of his long-term EEG during acquisition.

Etiology of de novo aphasic SE

In a literature review of 12 cases with de novo aphasic SE, the patients had the following causes: ischemic (N = 5), bleeding (N = 2), brain tumor (N = 2), encephalitis (N = 1) or head trauma (N = 1) [3]. A later series reported on seven patients with aphasic SE; in four, the status was the first seizure. Among those, three had recent or chronic vascular lesions, and one had a systemic infection [8]. The aforementioned retrospective study reported on 28 aphasic SE patients; eight of which had de novo SE [10]. In these

eight patients, etiologies included glioblastoma, subarachnoid hemorrhage, subdural hemorrhage, cerebral venous thrombosis, hyperglycemia, stroke, autoimmune encephalitis, unknown.

Differential diagnoses

Aphasia may occur as a postictal phenomenon. Postictal language delay in one study lasted for a maximum duration of 21 min [15].

Repeat seizures have been discussed as a differential diagnosis in the literature. According to Gastaut et al., recurrent focal EEG seizure patterns without interictal clinical symptoms should not be classified as SE [16]. Dinner et al. reported that the persistence of aphasia between EEG seizures is essential for the diagnosis of aphasic SE [17]. These authors considered two possible reasons for this phenomenon: an underlying structural lesion or postictal Todd paralysis involving the language area. The latter theory is supported by Dinner's case; that there is a clinical correlation between the number of seizure patterns and the degree of aphasia. In our case, one could also consider repetitive seizures with aphasia in between. Since the aphasia was stable over a period of seven months without clinical fluctuation, we conclude that our patient did not have repetitive seizures with postictal aphasia. EEG showed left temporal to posterior rhythmic theta activity as a seizure pattern, which correlated with aphasia, while vigilance was maintained. The co-occurrence with frequent ictal seizure patterns in a matching localization and the resolution of aphasia with less frequent such patterns leads to the conclusion that the aphasia was ictal.

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**Fig. 2.** Continuous long-term electroencephalography section. It shows a typical ictal alpha pattern arising from slowed activity in the left posterior temporal contacts (arrow 1). After 26 s, the rhythmic activity propagated to the left centro-parietal contacts (arrow 2). The rhythmic activity ended 80 s after onset (arrows 3, 4). Note: electrodes are named according to the American system.

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#### Table 1

Rosenbaum's criteria for aphasic status epilepticus, modified by Grimes and Guberman [3]. All five criteria need to be fulfilled.

- 1. The patient must have language production during the seizures.
- 2. Language production must show aphasic features.
- 3. Consciousness must be preserved.
- 4. The seizures must be correlated with the aphasia, as documented by EEG monitoring and behavioral testing.
- 5. The aphasia should resolve, or nearly so, concurrent with successful treatment of the seizures.

Abbreviations: EEG, electroencephalography.

### Treatment

Refractory NCSE usually requires intravenous application of ASM [18]. The present patient did not respond to oral ASM triple therapy plus a benzodiazepine, but responded upon intravenous administration of phenytoin. The response was maintained, despite discontinuation of two standard ASM and the benzodiazepine. Phenytoin was changed to oral administration after substantial improvement.

### Limitations

This report has limitations. It deals with a single case only and approaches this in a retrospective manner. The EEGs and the clinical investigations did not followed according to a standardized scheme. Therefore, all these aspects restrict the generalizability of our observation.

# Conclusions

As aphasic SE is a rare phenomenon with a broad differential diagnosis, it may easily be overlooked leading to a delayed diagnosis. The present case demonstrates that some patients with aphasic NCSE have a protracted course and require long-term EEG for diagnostic certainty which may be, supported by DWI sequences on brain MRI. Like other forms of SE, it is often necessary to administer intravenous ASM to terminate ongoing seizures.

# **Ethical statement**

The patient gave written informed consent for publication of this report and associated figures. (Please note that this retrospective report would be exempt from the obligation to obtain informed consent for publication by the Health Data Protection Act North-Rhine Westphalia of 22 February 1994. According to this act, research personnel may use for purposes of scientific research patient data to which they already have access due to their work in a hospital without consent of the patient.)

### **Declaration of Competing Interest**

Christian Brandt has received personal compensation from Angelini Pharma/Arvelle Therapeutics, Desitin, Eisai, GW Pharmaceuticals, UCB Pharma, and Zogenix for consulting services or speaking activities.

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The other authors have nothing to declare.

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