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CASE STUDY

Negative seizures

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Abstract

Negative phenomena can occur with seizures, but some ictal negative manifestations are rare and may lead to misdiagnosis. A patient series is presented with unusual ictal negative phenomena: neglect syndrome, catastrophic depression, apraxia, aphasia, amnesia, homonomous hemianopsia, and hemiparesis. One had repeated episodes with PLEDs but no parenchymal lesions. Clinicians should consider seizures in the setting of unexplained deficits, even if there are no positive ictal phenomena. (*JINS*, 2000, *6*, 731–733.)

Keywords: Seizures, Aphasia, Neglect

INTRODUCTION

Common negative ictal phenomena include loss of awareness or muscle tone (Wyllie, 1997). However, other types of negative ictal manifestations are rare, and the underlying epileptic mechanism is frequently not appreciated.

CASE REPORTS

Case #1

A 43-year-old right-handed man who was 7 years post-right-temporal-lobectomy had persistent seizures with a recent increase in frequency. He had numerous daily seizures described as left facial tingling and twitching, which then extended to involve the left arm and occasionally secondarily generalized. Interictal neurological examination was normal except for mild memory disorder. Multiple seizures with right centroparietal onsets were recorded on video EEG. The patient was not aware of his seizures when they remained localized to the right centroparietal region. During several of these recorded seizures, the patient was examined by a behavioral neurologist. With each seizure, the patient had a left arm pronator drift, asterixis, and neglect syndrome. EMG recordings demonstrated that left arm asterixis was temporally related to the rhythmic epileptic discharges on EEG. Ictal neglect was evidenced by extinction to double simultaneous stimuli in the visual and tactile modalities, line bisection to the right, hemi-inattention on cancellation tasks, hemidyslexia, hemispatial neglect in drawings, and denial of weakness or other deficits (see Figures 1–3).

Case #2

A 52-year-old right-handed man with intractable complex partial seizures since childhood underwent evaluation for epilepsy surgery. Interictal neurological examination was normal. Interictal scalp EEG revealed left temporal epileptiform discharges, and ictal EEG was poorly localized with bilateral (L > R) posterior cerebral onset. A small left occipital cortical dysplasia was present on MRI. Left occipitotemporal depth and left occipital subdural electrodes were implanted revealing multiple seizure onsets in the left occipital associative cortex. During some seizures, a right homonomous hemianopsia was demonstrated by confrontational testing of the patient's visual fields. However, the patient had no postictal recall of the visual deficits or other events that occurred during the seizure.

Case #3

A previously healthy 42-year-old right-handed man presented with a 10-day history of progressively increasing bifrontal headaches and visual spells, which were initially swirling colors but now were blurred or loss of vision in the right visual field. MRI revealed recent left occipital infarc-

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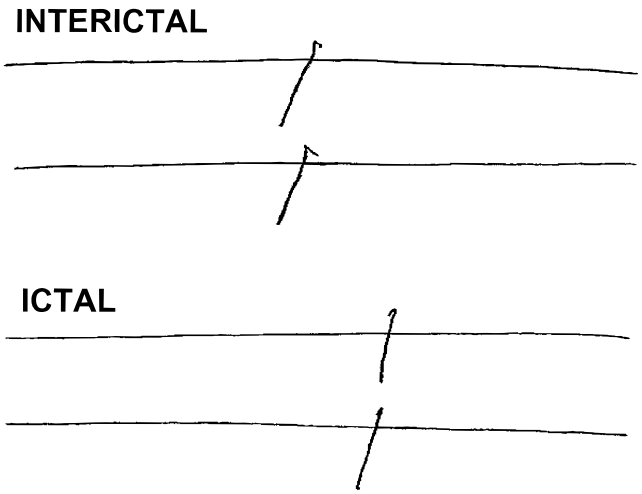

Fig. 1. Line bisections during interictal and ictal conditions.

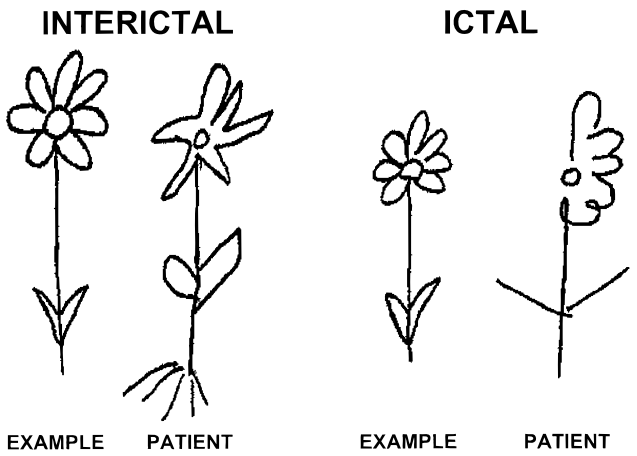

Fig. 3. Drawings of flower during interictal and ictal conditions.

tion with petechial hemorrhages. Angiography was normal, and no embolic source was identified, but the patient was discovered to have insulin-dependent diabetes. Interictal neurological examination was normal. During video EEG, visual loss was associated with 12- to 15-Hz epileptiform activity in the left occipital lobe. The spells resolved with phenytoin.

Case #4

A 61-year-old right-handed male physician suffered left MCA embolic infarction, secondary to intermittent atrial fibrillation. His deficits resolved over several months, except for mild residual word finding difficulties and rare paraphasic errors. The next year, he had onset of multiple episodes of right hemiparesis, clear ideomotor apraxia, and increased dysphasia lasting minutes to hours. Initially misdiagnosed as transient ischemic attacks, the spells continued despite adjustment of coumadin. EEG during one episode revealed left cerebral slowing and repetitive epileptic discharges from the left temporal region. Neurological exam during this episode confirmed the above-noted deficits. MRI showed no extension of his prior infarction. The patient was treated with phenytoin and had no recurrences of his episodic deficits.

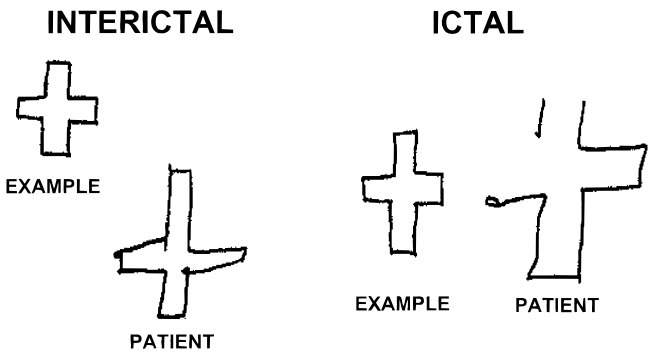

Fig. 2. Cross drawings during interictal and ictal conditions.

Case #5

A 48-year-old right-handed woman with metastatic breast cancer involving the skull underwent radiation therapy. She had a generalized tonic clonic seizure, and prolonged "postictal confusion." On neurological examination, she was alert and responsive but had global aphasia, ideomotor apraxia, right homonomous hemianopsia, right lower facial weakness, right hemiparesis, slight right-sided tendon reflex preponderance, and equivocal right Babinski. She also exhibited a weeping depressed affect. MRI with and without gadolinium showed multiple skull mets, but no parenchymal lesions. CSF was normal. The patient was treated with phenytoin and markedly improved over several days and then had further slow resolution of all signs and symptoms over 3 weeks. One month later, the patient had sudden onset of loss of awareness with lip smacking and right-sided weakness. She was again found to have aphasia, apraxia, right hemiparesis, and severely depressed affect. Transient focal clonic activity in the right upper extremity was seen despite a phenytoin level of 18 μg/ml. EEG revealed periodic lateralized epileptiform discharges (PLEDs) in the left parietal region. The patient was given lorazepam 2 mg IV, which reduced her epileptic discharges and produced some improvement in her aphasia and right hemiparesis. Phenobarbital was added to her drug regimen, and a major improvement in her aphasia and weakness were seen over the next several days. Concomitantly, there was a dramatic disappearance of her catastrophic depression. Complete resolution of her neurological deficits occurred over 2 weeks. Repeated MRIs with and without gadolinium again showed no parenchymal lesions.

DISCUSSION

Only one prior report of ictal neglect exists (Heilman & Howell, 1980). In that case, the patient had ictal tactile ex-

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tinction for left-hand stimuli, and had line bisection to the far left during focal right parieto-occipital seizures. In the immediate postictal period, he bisected lines to the far right. Our patient (Case 1) exhibited extinction in both the tactile and visual modalities, but bisected lines to the right during his seizure. In addition, we also demonstrated that he had other components of neglect including hemi-inattention on cancellation tasks, hemidyslexia, hemispatial neglect in drawings, and denial of weakness or other deficits. He did not have asomatognosia, although this has been described with seizures (Feinberg, et al., 1998). His ictal asterixis has also been called negative myoclonus (Tassinari et al., 1998) and is likely related to the same mechanisms underlying negative motor effects seen with cortical electrical stimulation in humans (Luders et al., 1995).

Cases 2, 3, and 5 had ictal homonomous hemianopsia; Cases 1, 4, and 5 had ictal hemiparesis; and Case 2 had ictal amnesia. Although rare, visual loss, somatic inhibition and isolated amnesia have been reported on occasion as ictal phenomena (Lee et al., 1990; Meador et al., 1985; Oliver et al., 1982).

Two patients (Cases 4 & 5) had ictal aphasia and apraxia. The deficits were linked to intermittent partial seizures in Case 4 and to PLEDs in Case 5. PLEDs are usually associated with an acute lesion such as an infarction; however, our patient never exhibited any parenchymal brain lesions on repeated MRIs, despite two distinct episodes. Further, her deficits partially resolved with lorazepam acutely, suggesting that the deficits were at least partially the direct result of epileptic activity, although postictal effects probably contributed to the slower complete resolution of deficits. Ictal aphasia has been reported (Ardila et al., 1988; Gilmore et al., 1981), but to our knowledge, clear demonstration of ictal ideomotor apraxia has not been described. Another interesting feature of Case 5 is the acute catastrophic depression, which resolved dramatically over the course of a few days in association with marked improvement in her aphasia and hemiparesis. Although patients with epilepsy have increased risk of mood disorders, depressed mood is rarely an ictal event (Wyllie, 1997).

In conclusion, focal seizures may inhibit normal neural function and produce a variety of negative phenomena that are topographically related to the site of ictal activity. Similar to negative phenomena produced by direct electrical stimulation, negative seizures appear more likely to occur with involvement of cortical associative cortex as opposed to primary sensorimotor cortex (Luders et al., 1995). Our first 2 cases had known longstanding epilepsy, but their ictal deficits were not appreciated prior to video EEG. The last 3 cases were initially misdiagnosed. The clinician should be aware of the occurrence of these rarely reported negative ictal phenomena, and consider them in their differential diagnosis. Given the number of cases that we observed in a relatively short period, such negative ictal phenomena may be more common than appreciated. However, a clinical diagnosis of negative ictal phenomena should be made with caution if not supported by EEG evidence of epileptiform activity.

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