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Unilateral Eye Blinking Arising From the Ictal Ipsilateral Occipital Area

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Abstract

We report on an 18-month-old boy with unilateral left eye blinking as a single ictal manifestation without facial twitching. The clinical onset of this phenomenon was first recorded (as an occasional event) at age 3 months, and it was overlooked. By age 6 months, the child's blinking increased to almost daily occurrence in clusters: during blinking the infant showed intact awareness and occasional jerks in the upper limbs and right leg. A video-electroencephalography (video-EEG) documented clinical correlation with a focal pattern arising from the left occipital region, and brain magnetic resonance imaging (MRI) revealed severe brain damage, consisting in poroencephalic hollows and increased spaces in the convexities involving a large area of the left cerebral hemisphere. The boy was prescribed sodium valproate (30 mg/kg/d), resulting in drastic reduction of his clinical seizures. Follow-up to his current age documented good general status, with persistent partial right hemilateral seizures. The blinking progressively disappeared, and is no longer recorded. The pathogenic hypotheses of the unilateral ictal blinking include involvement of the ipsilateral cerebral hemisphere and/or the cerebellar pathways. Review of previous reports of unilateral eye blinking, arising from the ictal ipsilateral brain, revealed that different damaged regions may give rise to blinking ictal phenomena, likely via the trigeminal fibres innervating the subdural intracranial structures and the pial vessels in the ipsilateral affected brain. The eye blinking in the present child represents a further example of an ictal phenomenon, which is predictive of the damaged brain region.

Keywords

Unilateral blinking, infantile spasms, brain damage, EEG anomalies, childhood

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Introduction

Pathological blinking may occur as a result of many conditions affecting the central nervous system, including epilepsy, stroke, and Parkinson's disease. *Contralateral blinking*, presenting with facial clonic twitching, is a frequent expression of partial epileptic attacks. Conversely, *unilateral blinking*, not associated with other facial twitching during an epileptic attack, has been rarely described. The triggering mechanism underlying this phenomenon is still unclear: many hypotheses have been raised, including involvement of the ipsilateral cerebral hemisphere or the ipsilateral cerebellar connections.[1]

We report an 18-month-old boy with left blinking as a single ictal manifestation. This phenomenon was correlated, by means of video-EEG recording, to a focal pattern involving the ipsilateral occipital area. Previous reports on children and infants with solitary, unilateral eye blinking arising from the ictal ipsilateral brain regions were reviewed.

Case Report

This 18-month-old boy was the first child of healthy, unrelated Italian parents. Pregnancy was complicated by hypertension by the third trimester. The child was the full-term product of a cesarean section. His weight at birth was 3850 g, his height was 50 cm, and his head circumference was 36 cm, all within the 50th percentile. APGAR scores were 5 and 7 at 1 and 5 minutes, respectively.

Since age 3 months, he showed occasional episodes of left eye blinking, which were overlooked by his parents. Over the

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following 3 months, blinking episodes increased to almost daily clusters. For this reason, he was referred to our outpatient clinic at the University Hospital in Catania. On admission, his body weight was 7100 g, his height was 68 cm, and his head circumference was 45 cm, all falling within normal limits. General examination was unrevealing. Neurological examination revealed partially unattained head control and right hemiparesis. The child was unable to maintain a sitting position constantly but was otherwise reactive to external stimuli. The anterior fontanel was open (2 × 2 cm across). Brisk tendon reflexes and bilateral ankle clonus were recorded. After admission to the ward, he presented several episodes of left eye blinking in clusters lasting a few seconds: These episodes were mostly single, but sometimes concurrent with subtle movements of the upper limbs and/or jerks of the right lower leg.

Routine laboratory investigations were normal. Fundus examination was normal, as were heart, abdominal, and pelvic ultrasonography. Serial EEG recordings showed (*a*) while awake, slow background activity, high-voltage spikes and polyspike and wave complexes over the left occipital areas and (*b*) during non–rapid eye movement sleep, spindles over the right central regions, polyspike and wave complexes, intermixed with short suppression bursts, more evident over the left hemisphere (Figure 1A and B). Video-EEG revealed, in awake status, a number of ictal events, consisting of blinking episodes at intervals of 10 to 30 seconds (average, 18 seconds), correlated to one another and associated with slow wave complex and superimposed fast activity followed by electric decrement (Figures 2 and 3). The left ictal blinking was often isolated, and did not involve the contralateral side of the face or the ipsilateral lower part of the face. During blinking, the infant showed awareness. The parents declined ocular electromyography of their child. MRI showed cortical and subcortical poroencephalic hollows in the left hemisphere, localized over the middle cerebral artery territory and involving the frontal–parietal and temporal areas. The space convexities were wider than normal, especially on the left side. These abnormalities were consistent with ischemic–hypoxic damage.

The boy was given sodium valproate (30 mg/kg/d), with drastic reduction of his blinking. Follow-up, at age 6, 12 and 18 months, showed his general condition was good. Blinking had progressively decreased to total disappearance, and he presented only partial right hemilateral seizures. After his last follow-up eye blinking was no longer recorded.

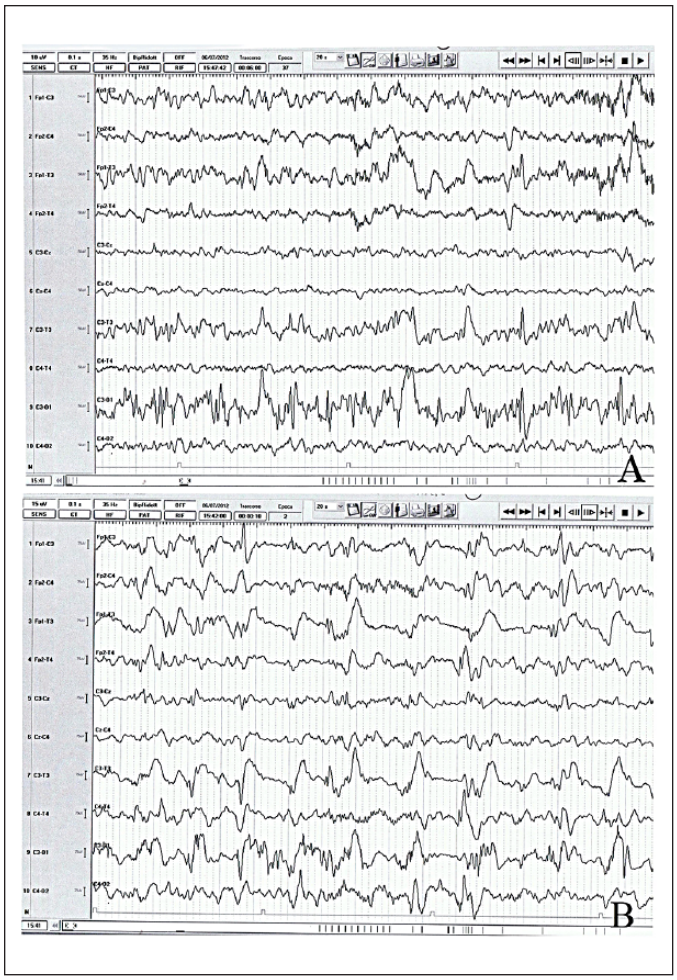

Figure 1. (A) Electroencephalogram recording during awake state shows slow background activity, high-voltage spikes, spike and polyspike and wave complexes over the left occipital regions. (B) Sleep EEG demonstrates the presence of spindles in the right central areas. Polyspike and wave complexes intermixed with short suppressions were recoded in the left hemisphere.

Discussion

The boy reported here had a spectrum of clinical and neurophysiologic features, which included (*a*) mostly single episodes of left-eye blinking, sporadically concurrent with clonic movements of the upper limbs, or jerks of the right lower leg; (*b*) high-voltage spikes and polyspike complexes prevalent in the left occipital, during interictal EEG recordings; (*c*) a pattern of slow-wave complexes, with fast activity, followed by electric decrement prevalently in the left occipital area, during ictal video-EEG recording; and (*d*) brain MRI involvement of the left frontal, temporal, and parietal areas. Blinking was the most apparent: Of note, it was unassociated with movements of other parts of the face. This child appeared to be in good condition. Apart from the epileptic attacks and the right hemiparesis, the child responded to external stimuli, and no dysmorphic signs or systemic anomalies were recorded.

The unilateral blinking was ictal, atypical, and overlooked. It is well-known that lateralized motor events may manifest during an epileptic attack with various clinical patterns: these events may have a practical, predictive role as indicators of the affected brain area. Head turning and tonic or dystonic posturing,[2,3] vomiting,[4] speech impairment,[2] and asymmetric limb position during an epileptic attack,[5] are highly useful indicators of the corresponding affected cerebral region. The case further

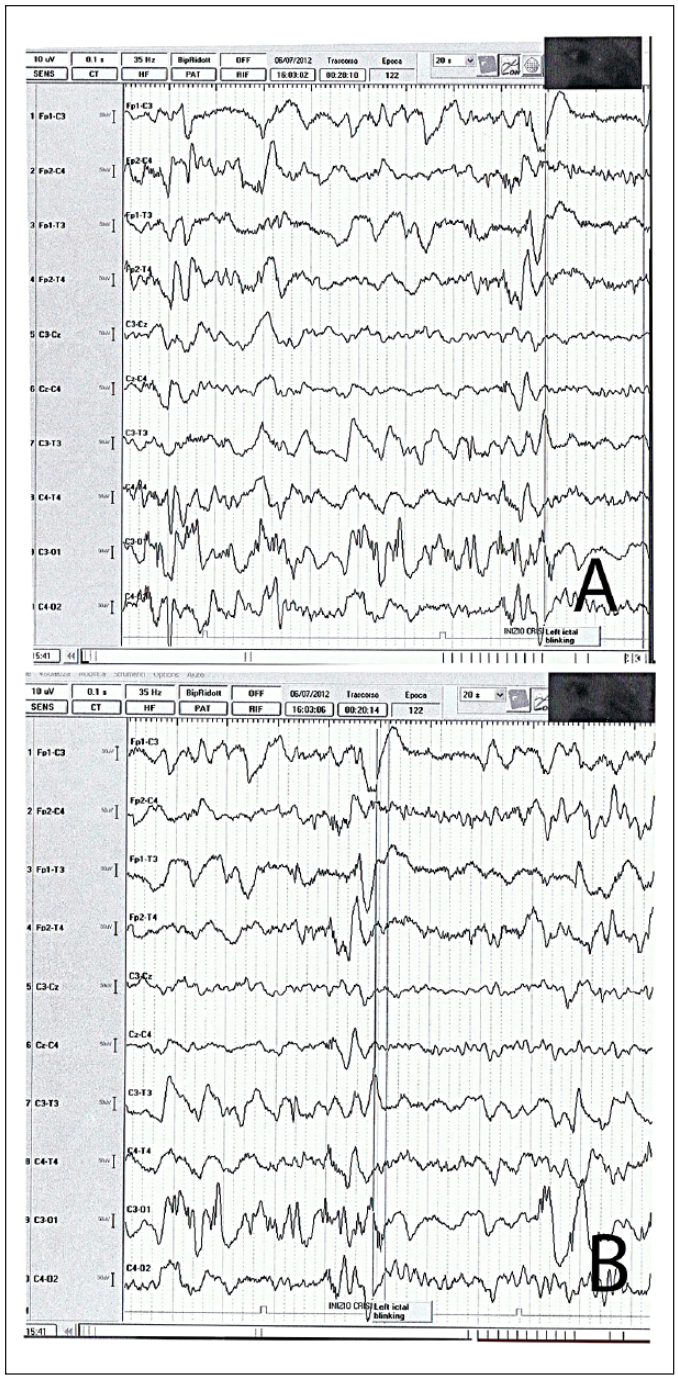

Figure 2. Video-EEG recording during awake state at eye opening (A) and closure (B) shows slow wave complex followed by fast activity and electric decrement.

expands this phenotype, confirming that lateralized blinking may be an indicator of the affected brain area.

Unilateral blinking, unassociated with clonic facial motor activity, has been rarely reported as an ictal event. Benbadis et al[6] described 14 patients who had unilateral eye blinking as an ictal component of their clinical spectrum of seizures. Although these authors[6] were unable to accurately localize the triggering area responsible for this peculiar blinking, they postulated that it was likely in the frontal region. Blinking epilepsy or seizures have been correlated both to temporal lobe dysfunction (demonstrated by subdural cortical stimulation)[7] and to temporal and extratemporal malfunctioning.[8] Pestana and Gupta[9] reported on a patient with ipsilateral blinking seizures, recorded by video-EEG, and a left frontotemporal ictal pattern on scalp EEG. They assumed that the ipsilateral blinking could have its origin in the trigeminal fibres innervating the subdural intracranial structures and the pial vessels in the ipsilateral frontotemporal regions,[9] as also shown by facial motor responses evoked by direct electric stimulation of the trigeminal root.[10] O'Connor and van der Kooy[11] showed that individual cells of the ophthalmic division of the trigeminal nerve diffusely innervate several intracranial structures (including the arteries and the dura), but separate cells in the ophthalmic division innervate extracranial targets. According to these authors,[11] ipsilateral blinking was obtained only when the trigeminal fibres were directly stimulated intracranially, whereas extracranial stimulation of the trigeminal fibres in the cornea or in the supraorbital region produced only the classical "blink reflex."

In this infant, video-EEG registration demonstrated that the unilateral ictal blinking originated from the ipsilateral occipital regions. Possible involvement of the ipsilateral cerebellum, as the area from which the ictal blinking originates, has been postulated in an infant with left cerebellar ganglioblastoma,[1] in patients with cerebellar damage,[12] and in trigeminal sensory transmission to the cerebellum, as demonstrated by fMRI.[13] Involvement of, and possible connections between, the temporal lobe and the cerebellar hemisphere has been reported as the most likely site of origin.[14]

Scalp EEG is not the gold standard in the localization of seizures, but it is very helpful for the overall assessment coupled with other modalities, and it is also useful in decision making for intracranial monitoring, which remains the gold standard. Clearly, a bilateral electromyography examination of the orbicularis oculi muscles would better define whether the blinking is synchronized with the seizures (as recorded at EEG) and that such phenomenon is a real unilateral movement. The present child's parents refused to perform this examination in their child.

The unilateral ictal blinking recorded in this child could represent an interesting observation. Unilateral eye blinking (without facial twitching) may represent an isolated expression of focal epileptic attacks. According to video-EEG registration in this boy, as previously recorded in the patient reported by Pestana and Gupta,[9] there is evidence that single unilateral blinking may be expression of an ipsilateral epileptic seizure, and that this seizure may originate from different areas of the affected cerebral hemisphere.

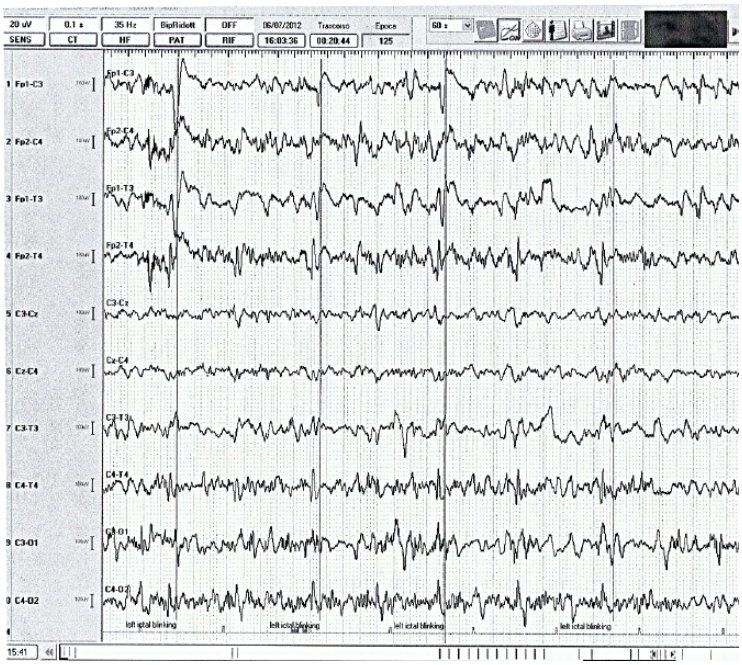

Figure 3. Video-EEG recording shows a "collage" of 4 ictal episodes distanced a few seconds from each other (at different times).

Declaration of Conflicting Interests

The authors declared no conflicts of interest with respect to the research, authorship, and/or publication of this article.

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