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LETTER

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Extreme delta brush in anti-NMDAR encephalitis: Mimics and chameleons

To the Editors,

We read with great interest the article published by Huang et al[1]. The authors presented novel electroencephalography (EEG) findings: spiky beta bursts that preceded generalized synchronous delta rhythms as ictal activity during orofacial dyskinesia in patients with anti-N-methyl D-aspartate receptor (NMDAR) encephalitis. As the EEG findings were different from so-called extreme delta brush (EDB), which is a specific marker of anti-NMDAR encephalitis,[2] they referred to it as an extreme delta variant (EDV). Although the authors

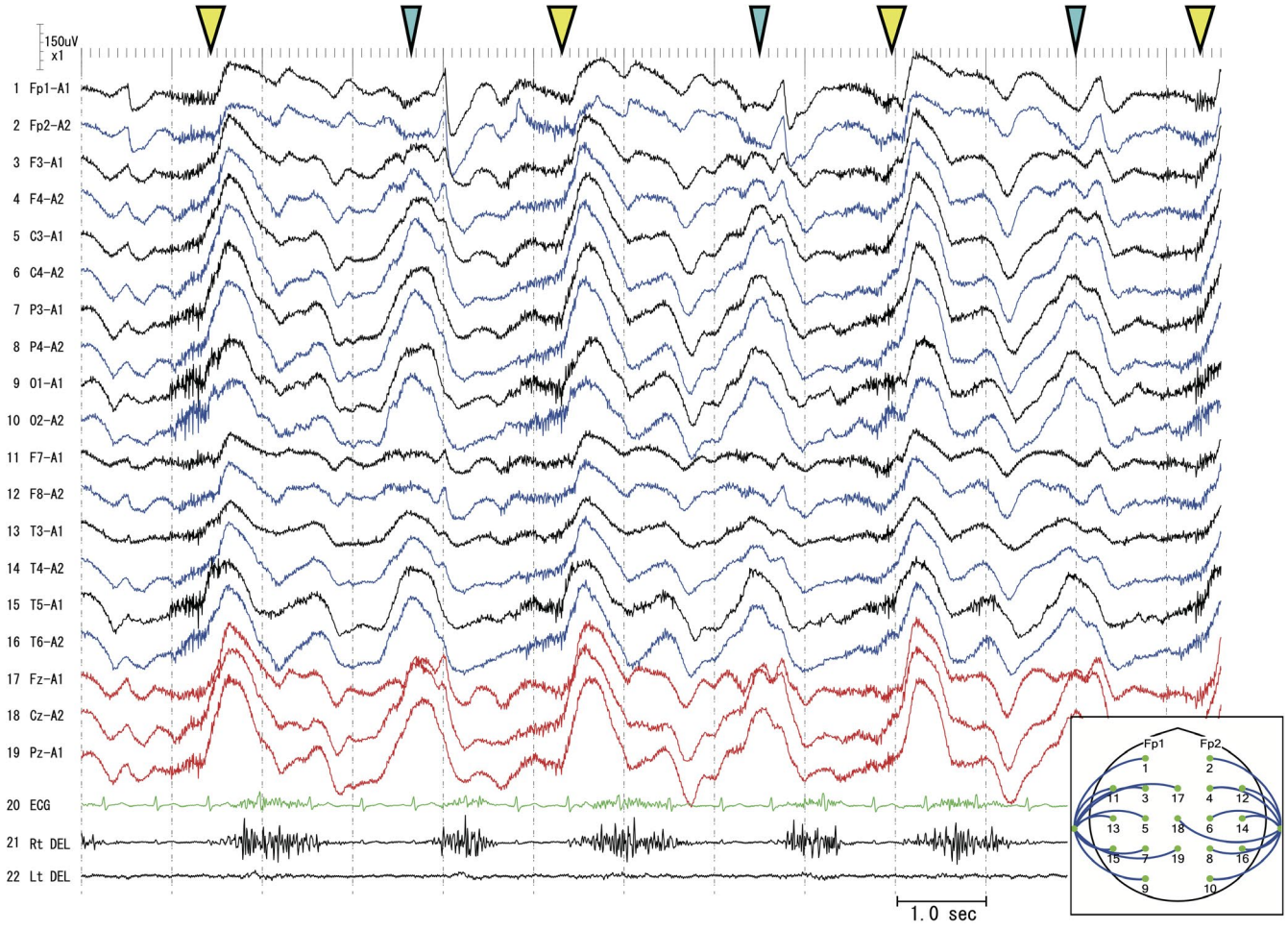

FIGURE 1 EEG and EMG (monopolar montage). EEG shows generalized rhythmic or semirhythmic delta activity that is most prominent in the frontocentral region. Beta bursts overriding on the delta waves are visible only when the orofacial dyskinesia involved facial muscle contractions (yellow arrowheads) but absent when the dyskinesia was mild (blue arrowheads). The beta bursts were not synchronized with EMG activity from the deltoid (DEL) muscles

reported that the EDV was sharply different from electromyography (EMG) artifacts, the spiky beta bursts may have contained EMG activity to some extent given several possible reasons. First, the amplitude of the beta burst was excessively large as the cortical activity in the scalp-recorded EEG. Second, although the distribution of beta bursts was generalized, the beta amplitude was lowest in the Cz. This finding was reasonable for EMG, given that the vertex region lacks muscles. To clarify the origin of the EMG artifact, a coronal bipolar montage (also known as a transverse montage) would have been helpful. Third, because the patients exhibited orofacial dyskinesia, which can involve several muscles (tongue, masseter, frontalis, and temporal muscles), the widely distributed synchronous beta might have been acceptable for EMG. As the true EDB was not associated with the presence of involuntary movements including orofacial dyskinesia,[3] the contamination of EMG artifacts could have been feasible in the present EDV (however, it should be also noted that a case of NMDAR encephalitis who showed EDB with choreoathetoid movements was reported).[4]

We herein present a 31-year-old woman with anti-NMDAR encephalitis, in whom EEG revealed an "EDB mimic." While involuntary orofacial and extremity movements were retained following initial anti-immune therapy, EEG revealed generalized high-amplitude rhythmic/semirhythmic 1–2-Hz slow waves (Figure 1). The delta waves were synchronous to the orofacial dyskinesia but asynchronous to the limb movements. Additionally, the delta waves were preceded by bursts of beta activity. However, the beta bursts were only visible when the oral dyskinesia involved the facial muscle contractions (yellow arrowheads). In contrast, when the oral dyskinesia was mild (not involving facial muscle contractions), bursts of beta activity were absent (blue arrowheads). A transverse montage was also helpful to clarify that the origin of the EMG artifact was prominent in the bilateral temporal regions (Figure 2). After sedation with intravenous midazolam, the dyskinesia disappeared along with the beta bursts (Figure 3A), and the generalized high-amplitude waves gradually became less noticeable (Figure 3B). The present EEG abnormality was sharply different from EDB in terms of the location of the beta activity. True EDB typically involves beta activity on the descending part of the delta slow.[2,5,6] In contrast, the present "EDB mimic" involved beta activity on the ascending part of the slow. Therefore, we concluded that the present EEG findings could include a combination of 1) EMG activities of the orofacial muscles and 2) large potentials of the genioglossus

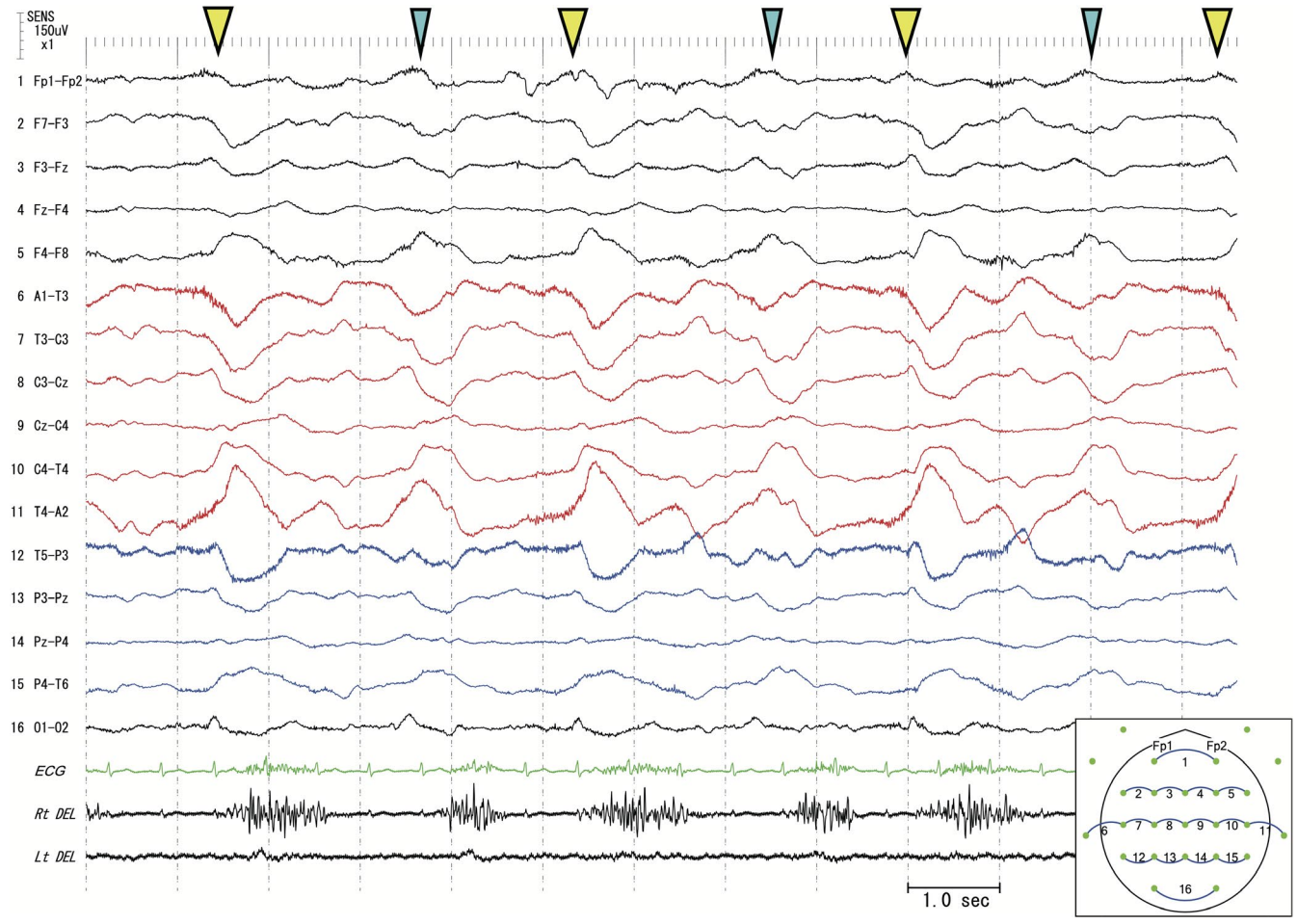

FIGURE 2 EEG and EMG (transverse bipolar montage). The time window of the EEG is the same as in Figure 1

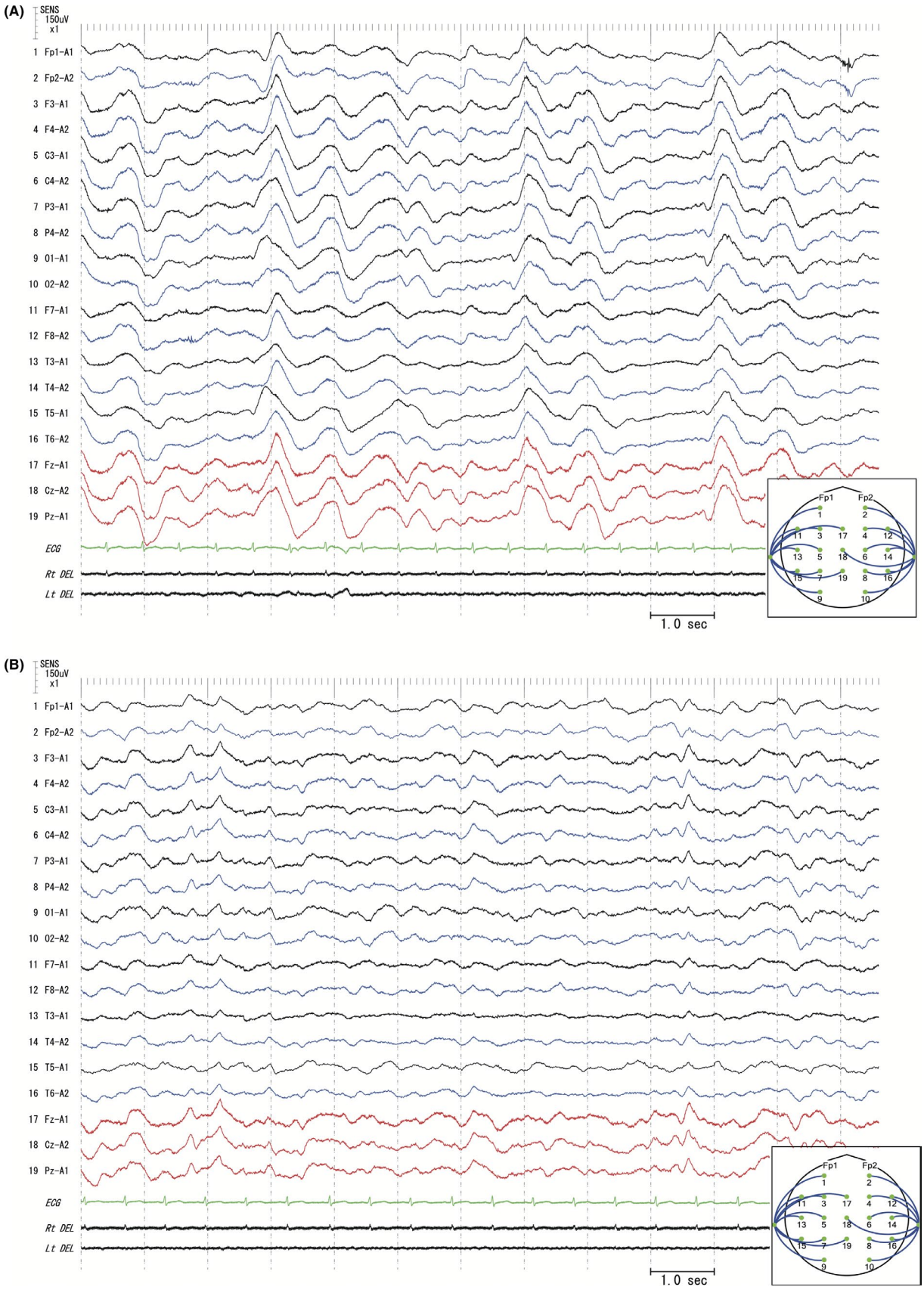

FIGURE 3 EEG and EMG after the sedation. The EDB mimic gradually becomes less noticeable after 1 min (A) and 10 min (B) from the initiation of intravenous midazolam injection

muscle (glossokinetic potential).[7] Since patients with anti-NMDAR encephalitis often exhibit orofacial dyskinesia, EDB mimics should be acknowledged.

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CONFLICT OF INTEREST

On behalf of all the authors, the corresponding author states that there are no conflicts of interest to disclose. We confirm that we have read the journal's position on issues involved in ethical publication and affirm that the present report is consistent with those guidelines.

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REFERENCES

1. Huang Q, Liao Y, Ma M, Wu Y. Delta brush variant: a novel ictal EEG pattern in anti-NMDAR encephalitis. Epilepsia Open. 2020;5:507–13.
2. Schmitt SE, Pargeon K, Frechette ES, Hirsch LJ, Dalmau J, Friedman D. Extreme delta brush: a unique EEG pattern in adults with anti-NMDA receptor encephalitis. Neurology. 2012;79:1094–100.
3. Gillinder L, Warren N, Hartel G, Dionisio S, O'Gorman C. EEG findings in NMDA encephalitis - A systematic review. Seizure. 2019;65:20–4.
4. Herlopian A, Rosenthal ES, Chu CJ, Cole AJ, Struck AF. Extreme delta brush evolving into status epilepticus in a patient with anti-NMDA encephalitis. Epilepsy Behav Case Rep. 2017;7:69–71.
5. Veciana M, Becerra JL, Fossas P, Muriana D, Sansa G, Santamarina E, et al EEG extreme delta brush: an ictal pattern in patients with anti-NMDA receptor encephalitis. Epilepsy Behav. 2015;49:280–5.
6. Zhang Y, Liu G, Jiang MD, Li LP, Su YY. Analysis of electroencephalogram characteristics of anti-NMDA receptor encephalitis patients in China. Clin Neurophysiol. 2017;128:1227–33.
7. Ayse KB, Dennis S, Rafeed A. Glossokinetic potentials: insights from intracranial EEG. Neurology. 2019;92:4.5-014.