■ Electroclinical reasoning report

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Epileptic Disorders

Ictal head roll: a seizure semiology from the anterior prefrontal lobe

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ABSTRACT

Longstanding epilepsy can lead to modulation of cortical networks over time and unexpected seizure onset zones. Frontal lobe seizures, in particular, can have diverse semiologies and evolution patterns. We present a male patient with drug-resistant epilepsy secondary to severe traumatic brain injury who underwent bilateral stereo electroencephalography (SEEG) for surgical planning. SEEG localized an ictal circular head roll to the right anterior prefrontal region. This was followed by spread to the left orbitofrontal region and later the left amygdala and hippocampus, at which point a different semiology with behavioral arrest, lip smacking and oral automatisms began. This case, in which an ictal circular head roll was localized to the anterior prefrontal region, demonstrates the complexity of broad seizure networks that develop over time, leading to remote seizure spread.

Key words: drug-resistant epilepsy, stereoelectroencephalography (SEEG), ictal circular head, anterior prefrontal region

VIDEO ONLINE

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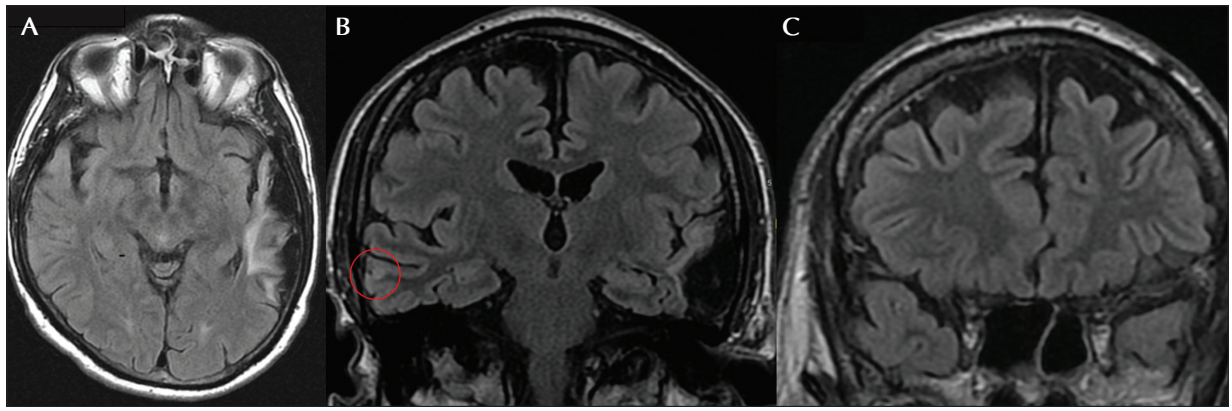
The goal of surgery in patients with drug-resistant epilepsy is to successfully identify the epileptogenic zone using ictal semiology and additional tools such as clinical history, neuropsychology, imaging, and electroencephalography [1]. Frontal lobe epilepsy poses a unique challenge as frontal ictal semiology can be heterogeneous and localization can be challenging due to the frontal lobe's large cortical volume and widespread connections [2]. We illustrate the case of a patient with an ictal circular head roll as an unusual semiology arising from the right anterior prefrontal lobe with consistent subsequent involvement of the contralateral mesial temporal region.

Case study

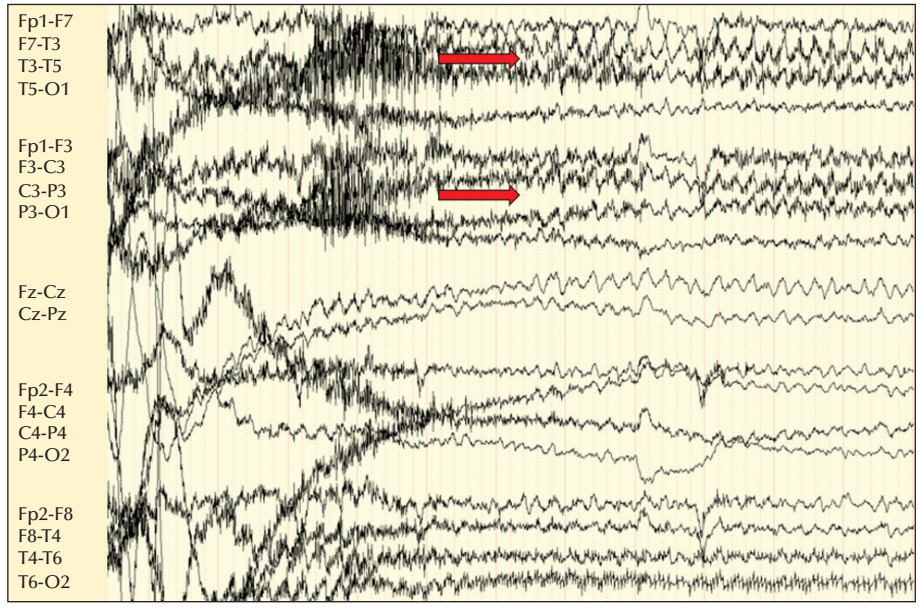
Clinical history and semiology

A 67-year-old, right-handed male with a past medical history of traumatic brain injury (TBI) at age 19 after a fall from an elevated height, with subsequent predominantly left frontotemporal encephalomalacia (*figure 1*), depression, and mild cognitive dysfunction, presented to our center for epilepsy surgery evaluation. He had drug-resistant epilepsy for more than 40 years, having failed at least seven different antiseizure medications (carbamazepine, clonazepam, gabapentin, lamotrigine, levetiracetam, oxcarbazepine, phenytoin, and

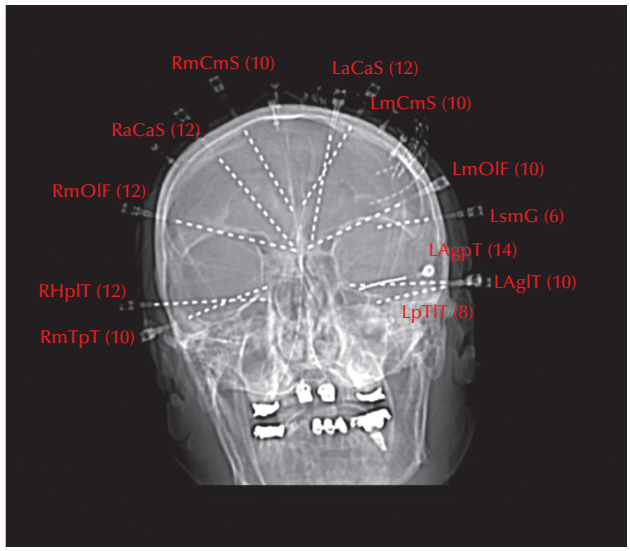


■ **Figure 1.** Brain MRI images. (A) Axial T2/FLAIR image showing left temporal encephalomalacia. (B) Coronal T2/FLAIR image again showing left temporal encephalomalacia as well as non-specific, mildly increased FLAIR signal within the right middle temporal gyrus (red circle). (C) Coronal T2/FLAIR image showing bilateral orbitofrontal lobes without any clear pathology.

cannabidiol) over the past 10 years. The patient and his partner described two different types of focal impaired aware (FIA) seizures. In the first, less frequent type, he would have a *déjà-vu* feeling, as if he was "going into a tunnel" followed by behavioral arrest. His second, and more frequent seizure type began with a circular head rolling motion, followed by behavioral arrest and lip smacking. Prior to starting medical therapy, the patient would have nocturnal focal to bilateral tonic-clonic seizures (FTBTC) as well. He was currently maintained on oxcarbazepine at 600 mg twice daily and clonazepam at 0.5 mg in the morning and 2 mg at bedtime. Despite these medications, he continued to have seizures nearly every other day. The patient underwent an ambulatory EEG showing bilateral independent anterior temporal sharp waves, maximal at F7 and at F8. One non-convulsive seizure of unclear onset but with better evolution over the left anterior quadrant was captured (*figure 2*). Neuropsychological testing indicated frontal and mesial temporal dysfunction that



■ **Figure 2.** Scalp EEG. Non-convulsive seizure with unclear onset but maximal evolution over the left anterior quadrant where rhythmic theta is seen (red arrows). Bipolar montage: left temporal chain, left parasagittal chain, central, right parasagittal chain, right temporal chain.



■ **Figure 3.** Location of SEEG electrodes on scalp and MRI. L: left; R: right; mOlF: mesial orbitofrontal to lateral frontal; aCaS: anterior cingulate to anterior superior frontal gyrus; mCmS: mid-cingulate to middle superior frontal gyrus; AgpT: amygdala to posterior temporal; AglT: amygdala to lateral temporal; pTlT: posterior temporal to lateral temporal; smG: supramarginal gyrus; HplT: hippocampal to lateral temporal; mTpT: mesial temporal to posterior temporal. The number in parenthesis after each abbreviation indicates the number of electrodes in each lead.

appeared more prominent over the left hemisphere, consistent with dominant hemispheric dysfunction, however, bilateral involvement was also suggested. Given the bilateral potentially epileptogenic foci seen on ambulatory EEG and long duration of epilepsy in a patient with traumatic brain injury, the patient underwent bilateral SEEG with frontal and temporal coverage (*figure 3*).

Results of SEEG

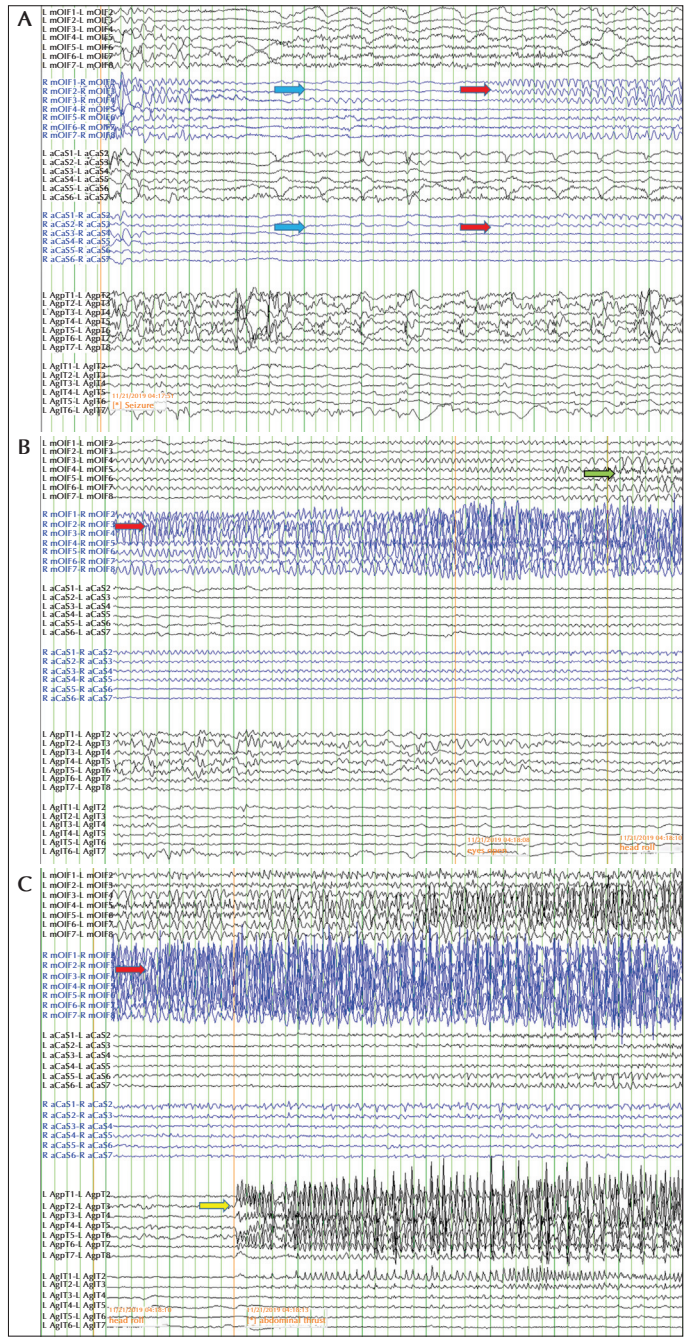
Seven seizures were captured, five originating from the right anterior prefrontal region and two originating from the left anterior prefrontal region. The anterior prefrontal region is composed of the mesial and lateral orbitofrontal electrodes as well as the anterior cingulate electrodes. Four of the five seizures originating from the right anterior prefrontal region had a clinical correlate, one was subclinical, and all the seizures from the left anterior prefrontal region were subclinical. The four clinical seizures started maximally in the right mesial orbitofrontal region and extended to the lateral frontal region, anterior cingulate and superior frontal contacts. This activity was electrographically characterized as a two-second burst of slow waves over this region followed by focal attenuation and gamma activity, which evolved into rhythmic alpha spikes maximal over the right orbitofrontal, lateral frontal and anterior cingulate contacts. This activity then spread to the contralateral left mesial orbitofrontal contacts, before spreading more diffusely throughout the lateral frontal lobe. About 15 seconds later, there was abrupt onset of rhythmic high-amplitude fast activity in the left amygdala (*figure 4A-C*).

The patient's maximal ictal activity was seen over the right anterior prefrontal region, most notably over the region spreading from the right mesial orbitofrontal to lateral frontal region. This corresponded to a stereotyped circular head rolling motion (*figure 4B, C*). The head roll began with the first movement towards the left, then downward and around twice, lasting for about 5 seconds. This was then followed by a brief abdominal thrust (*video sequence 1*). About 10 seconds after the circular head roll, the patient displayed behavioral arrest, lip smacking and hand automatisms which correlated with spread to the left amygdala and hippocampus (*figure 4C, figure 5*). Subsequent cortical brain mapping revealed afterdischarges in the bilateral mesial orbitofrontal and right anterior cingulate regions, a "pre-seizure feeling" in the right anterior cingulate, and an electroclinical seizure manifesting only with an aura with direct 50-Hz stimulation of the left amygdala at the 2-mA stimulation level.

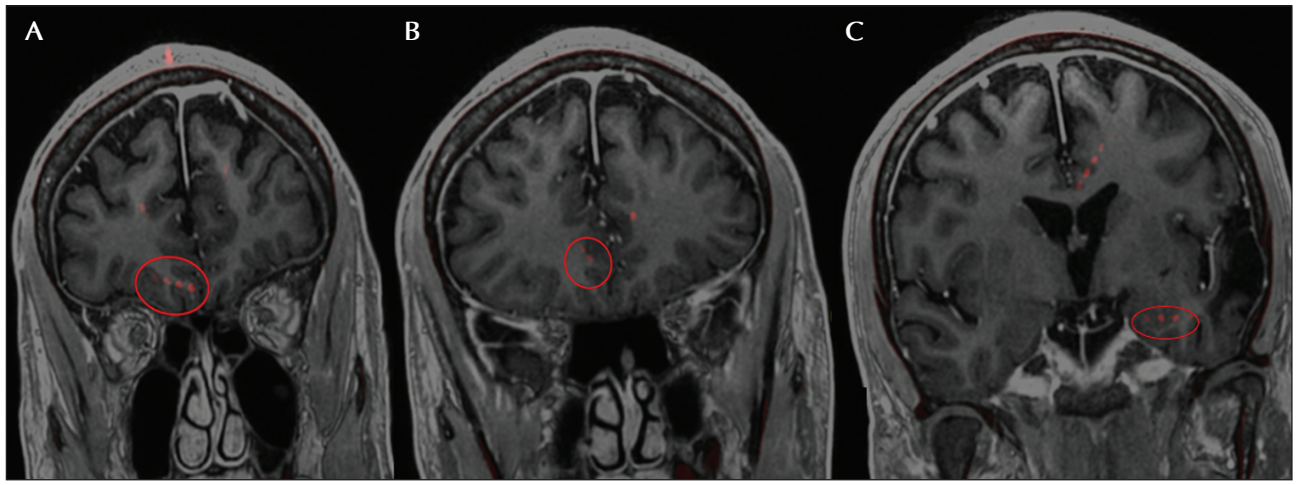
Analysis of anatomo-electro-clinical correlations

Frontal lobe seizures present with diverse clinical semiology, encompassing simple and complex motor behaviors. Electrographically, precise localization can be challenging given the brief time period from ictal appearance to clinical manifestations, as well as the wide rapid connections of the frontal lobe to other brain regions leading to rapid synchronization of electrographic activity [3, 4]. Furthermore, the frontal lobe encompasses a large area including buried portions in the mesial and orbitofrontal regions that are not easily accessible via scalp EEG; therefore, additional diagnostic tools such as ictal-SPECT and intracranial EEG are often used to further lateralize and localize frontal lobe seizures [3, 5].

Orbitofrontal epilepsy has traditionally been challenging to describe given its relative rarity and apparent heterogeneity of presentation [4, 6]. However, it may be suspected when the following criteria are met: onset during sleep, absent or non-specific cephalic auras, hypermotor manifestations, and/or an EEG



■ **Figure 4.** (A-C) SEEG traces. (A) Stereo-EEG showing seizure onset with slow waves in the right mesial orbitofrontal region (seen immediately after montage labeling), followed by diffuse attenuation with overlying gamma activity over the anterior prefrontal region (blue arrows) and then rhythmic alpha activity (red arrows). (B) Seizure evolution with rhythmic alpha activity over the right anterior prefrontal region, with highest amplitude over the right mesial orbitofrontal to lateral frontal area and lower amplitude over the right anterior cingulate area, that evolves to rhythmic beta activity (red arrow), corresponding clinically to a counterclockwise head roll. To a lesser extent, there is rhythmic alpha seen over the left mesial orbitofrontal to lateral frontal region (green arrow). (C) Continuation of the right anterior prefrontal seizure (red arrow) with new-onset independent rapid beta spiking in the L amygdala/ hippocampal region (yellow arrow). L: left; R: right; mOlF: mesial orbitofrontal to lateral frontal; aCaS: anterior cingulate to anterior superior frontal gyrus; AgpT: amygdala to posterior temporal; AglT: amygdala to lateral temporal.



■ **Figure 5.** Location of seizure onset on SEEG showing the origin of seizure in the right anterior prefrontal region, encompassing the right mesial orbitofrontal region, innermost four electrodes (RmOlF 1-4) (A) with a spread pattern to the left anterior cingulate region (RaCaS 1-3) (B), finally spreading to the left amygdala region (LAgpT 1-4) (C).

showing frontotemporal discharges that may or may not clearly lateralize according to a recent case series [6].

The heterogeneity of orbitofrontal seizures may be secondary, at least in part, to the vast connections of the orbitofrontal lobe to other brain regions, particularly the mesial and lateral frontal lobes and the limbic system. In one case series, eight patients with confirmed orbitofrontal epilepsy demonstrated electrical activity that invariably spread to other frontal or temporal lobe regions, with subsequent clinical symptoms reflecting the location of the spread pattern [7]. Orbitofrontal spread that involves the anterior cingulate region may display a seizure semiology of hypermotor behavior with or without emotional overtones, whereas mesial temporal lobe spread may involve sensations of *déjà-vu*, lip smacking and hand automatisms [4-7]. A recent orbitofrontal SEEG series further demonstrated that patients with orbitofrontal epilepsy propagating frontally tended to use the anterior cingulate network while the group moving temporally tended to spread via the amygdala connection [8]. This latter study also showed an association between orbitofrontal seizures with a "frontal" signature and anterior cingulate and anterior insula hypometabolism on FDG-PET, further suggesting these as possible propagation pathways. The spread patterns of orbitofrontal seizures were also studied in a larger frontal lobe SEEG series where the two most anterior and mesial groups demonstrated integrated gestural motor behavior (described as more natural and coordinated motor behaviors) and distal stereotypies [9]. These seizures tended to spread both ventrolaterally as well as to the anterior cingulate region, with the medial-most group spreading temporally via the amygdala [9]. Our case demonstrates a similar electrographic spread pattern involving the anterior prefrontal network from the mesial orbitofrontal region to the anterior cingulate, lateral frontal region, and subsequently contralateral amygdala. The ictal circular head rotation in our case appears natural as an integrated motor behavior, similar to what one might do prior to exercise or stretching. This ictal behavior occurs once the seizure has spread, to involve the anterior prefrontal cortex as seen by the SEEG contacts that span from the mesial orbitofrontal to lateral frontal and to a lesser extent in the anterior cingulate to superior frontal leads.

Ictal circular head rolls should be distinguished from ictal head turns which may be of a versive or non-versive type. Versive head turns, also known as tonic turns, are characterized by a sustained contraction of the head in a particular direction, often in an unnatural posture, as opposed to non-versive or non-tonic turns which are brief and may appear spontaneous or voluntary [10]. Prior studies looking at head turns in temporal and frontal lobe epilepsy have found that contralateral versive head turns, often with associated gaze deviation, always occur prior to secondary generalization of the focal seizure onset, as compared to ipsilateral head turning [11]. While versive head turning with its usual associated gaze deviation has been felt to indicate involvement of the frontal eye fields, our patient did not demonstrate either a fixed

versive head turn or gaze deviation during the head roll episodes.

In contrast, non-versive head turns often occur earlier during the seizure and are more likely to be ipsilateral to the seizure focus [10]. Ipsilateral head turns have further been noted to occur earlier during the seizure onset in patients with frontal lobe (with a median of 0.5 seconds) as opposed to temporal lobe epilepsy (with a mean of 16 seconds) [11]. This may be considered an element in our case, however, the initial head movement begins with a non-tonic left head turn (contralateral), immediately followed by circular head rotation back and toward the right (ipsilateral).

Our patient's circular head roll semiology should be contrasted with the rarely reported "gyratory seizures" which have been variably described as rotation around the body axis by 180 degrees [12]. Gyratory seizures originated more commonly in the frontal compared to the temporal lobes in a video EEG series [12]. In terms of directionality, for patients with gyratory seizures that did not begin with a head turn, the localization was ipsilateral to the seizure onset zone, while in those patients in whom there was a clear versive head turn that preceded the rotational movement, the onset was contralateral [12]. Two prior reports looking at rotational seizures along a body axis, while patients were standing, described both clockwise and counter-clockwise body rotations and have suggested a connection with the basal ganglia given that lesions to this region have been noted to cause circling movements in animals [13, 14].

While an intriguing parallel, our patient's head rolling motion was limited to the head and neck axis rather than the entire body axis, as described in the studies above, where patients additionally were often standing rather than laying down. Nevertheless, taking the prior literature into consideration, this suggests that ictal head rolls may be considered a form of complex anterior prefrontal automatism whose axis of rotation is ipsilateral to the seizure onset zone relative to the patient, and possibly contralateral when a versive head turn precedes the head roll. Given that our head turn always started with a left head movement before rightwards rotation, it may fit the semiology previously suggested for gyratory seizures [12]. Nevertheless, given the rarity of the head roll semiology, larger case series are needed to definitively characterize the localizing findings of this rotational semiology.

A final note to be considered is the pelvic thrust which immediately followed the two clockwise head rolls in our patient. Pelvic thrusts have been described in frontal lobe seizures including in a recent small SEEG series contrasting medial and lateral orbitofrontal epilepsy [15]. In this series, pelvic thrusting and pedaling were described to appear semiologically after seizures arising from the lateral orbitofrontal cortex propagated to the lateral frontal lobe [15]. This is seen in our case and the electrical discharges are seen to spread from the mesial orbitofrontal cortex to the lateral frontal lobe at the time of the brief abdominal thrust.

A surprising aspect of our case may be the confirmed seizure onset zone. Despite the prominent left temporal encephalomalacia seen on brain MRI, the patient's clinical seizures electrographically started in the right anterior prefrontal region. This seizure onset zone may be secondary to kindling of additional brain regions due to longstanding epilepsy, versus involvement of the right anterior prefrontal region in the setting of *coup-contrecoup* injury. *Coup-contrecoup* injuries affecting the anterior temporal and inferior frontal regions have been reported in traumatic brain injury, with common causes being falls from heights due to accelerating forces on impact, as our patient experienced [16, 17].

Another interesting aspect of our case concerns the unusual spread pattern from the right anterior prefrontal region towards the left amygdala as best seen originating on the SEEG traces from the mesial orbitofrontal to lateral frontal contacts. While orbitofrontal seizures are known to propagate into the temporal region, we suspect that the large area of encephalomalacia in the left temporal lobe, which was hyperexcitable as demonstrated by the abundant interictal discharges over this area, led to a preferential spread pattern towards the left instead of the right in this case. This is reinforced by the fact that the patient experienced an electrographic seizure with an associated aura when the left amygdala was directly simulated during brain mapping. The anterior insular region may have also been involved in this spread network from the orbitofrontal to the anterior temporal areas and was not explored in this SEEG study, which is a limitation of our study.

Results following surgery

The patient eventually underwent bilateral responsive neurostimulation (RNS System, Neuropace, Mountain View, CA) placement with a stimulating strip in the right mesial frontal to anterior cingulate region and a depth electrode along the long axis of the left amygdala and hippocampus. There was an additional strip electrode placed along the left mesial orbitofrontal region, yet it was not connected to the device. He has tolerated the procedure well without long-term complications. A detection change was recently made to his device and thus it is too early to determine seizure outcome at this time. The patient has recently reported that he will sometimes feel an aura that does not fully progress since making this change, however, at this time he remains with frequent ictal head rolls.

Conclusions

Frontal lobe seizures have diverse clinical and electrographic manifestations, and in many cases are intimately connected to electrographic networks involving the cingulate gyrus and mesial temporal lobe. Both longstanding epilepsy and traumatic brain injury, as in the case presented here, may alter neural networks and produce seizures from unsuspected places. Our case illustrates the importance of seizure semiology in predicting the epileptogenic zone and highlights the importance of not underestimating the evolution of epileptogenic networks that may develop over time. In our case, an ictal circular head roll localized to the right anterior prefrontal lobe on four separate occasions. An ictal head roll, an uncommon finding, may assist the neurologist in localizing seizures to the anterior prefrontal lobe when confronted with this rare semiology. ■

Supplementary material.
Summary slides accompanying the manuscript are available at www.epilepticdisorders.com.

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GT previously held shares in a diversified healthcare index-exchange traded fund unrelated to this study (2019). FP has consulted for Zimmer-Biomet and Neuropace. SG has received honoraria for educational activities on behalf of Monteris. NJ receives grant funding paid to her institution for grants unrelated to this work from NINDS (NIH U24NS107201, NIH IU54NS100064) and PCORI. She receives an honorarium for her work as an Associate Editor of *Epilepsia*. MF has received travel reimbursement for educational activities on behalf of Neuropace. DB and NL declare no conflicts of interest.

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Legend for video sequence

The head roll begins with the first movement towards the left, then downward and around twice, lasting for about 5 seconds. This is followed by a brief abdominal thrust. About 10 seconds after the circular head roll, the patient displays behavioral arrest, lip smacking and hand automatisms.

Key words for video research on www.epilepticdisorders.com

Phenomenology: focal automatisms seizure
Localisation: frontal lobe (right)
Syndrome: drug resistant focal epilepsy
Aetiology: post-traumatic

TEST YOURSELF

(1) What electroclinical features may help suggest a diagnosis of orbitofrontal epilepsy?

(2) How do head turns assist with lateralization of the seizure onset zone?

(3) Where do ictal head rolls localize to?

(4) What diagnostic tests may be used to further localize seizures arising from the orbitofrontal region, when scalp EEG and MRI fail to localize the ictal zone?

Note: Reading the manuscript provides an answer to all questions. Correct answers may be accessed on the website, www.epilepticdisorders.com.