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Localization value of ictal turning prone

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ABSTRACT

Purpose: Ictal semiology complements ictal EEG in identifying the likely epileptogenic zone. Ictal turning prone (ITP) with body turning of 90 ° or more can be seen with frontal lobe epilepsies. The aim of our study was to evaluate the localizing value of ITP in a general population of patients undergoing long term video-EEG monitoring.

Methods: We reviewed our epilepsy monitoring unit database for adult patients with recorded habitual seizures with ITP. All 16 patients identified had continuous video-EEG monitoring using standard scalp electrodes; eight patients also had intracranial EEG monitoring. We only included focal seizures without evolution to bilateral tonic-clonic activity.

Results: We identified 16 patients with ITP, mean age of 32.5 years (range 18–50). ITP was consistently seen in at least one focal impaired awareness seizure of all patients. Ictal onset zone on scalp EEG was left temporal in five, right temporal in three, left frontal convexity in two, right frontal convexity in two, probable right medial frontal in three and probable left medial frontal in one patient. Direction of ITP was uni-directional in 12 patients while 4 patients had ITP in opposite direction in different seizures.

Nine patients underwent epilepsy surgery; five patients had Engel class I outcome and four patients had Engel class III outcome.

Conclusions: Ictal turning prone does not have a consistent single localizing or lateralizing value and can be seen with various epileptogenic zones including medial frontal, lateral frontal or temporal. ITP direction can vary even with a single epileptogenic zone.

1. Introduction

Ictal turning prone (ITP) was first described by Saygi et al as an ictal feature of frontal lobe "focal impaired awareness seizures" that distinguished them from psychogenic nonepileptic seizures with otherwise similar semiology [1]. Ictal turning prone can be considered a variation on "ictal body turning along the horizontal axis" described by Leung et al as a sign that helped further localize frontal lobe focal impaired awareness seizures [2]. Ictal body turning along the horizontal axis was defined as "truncal turning without any tonic element of the extremities for 90° or more, parallel to the body axis and horizontally". It was found to favor mesial frontal localization in frontal lobe seizures with hypermotor semiology [2].

Frontal lobe epilepsy (FLE) is the second most common localization related or focal epilepsy and accounts for approximately 20–30 percent of all focal epilepsies [3]. Focal impaired awareness seizures in FLE are often difficult to diagnose on surface EEG [4–6], and are frequently misdiagnosed as non-epileptic events such as psychogenic seizures or parasomnias [1,7]. Hence, clinical semiology is important for diagnosis.

Having observed ITP in some patients with temporal lobe epilepsy we evaluated the localizing value of ITP in focal impaired awareness seizures in patients undergoing long-term video-EEG monitoring.

2. Methods

The Vanderbilt epilepsy monitoring unit database was reviewed for adult patients with recorded habitual seizures that included ITP over a period of 12 years (2004-15). We defined ITP as ictal body turning for 90 ° or more, along the longitudinal body axis, leading to a prone position in bed. Only patients who had ITP as an early feature during their seizure (within the first 20 s) were included. We excluded ITP driven by versive head turning; hence we included only focal seizures without evolution to bilateral tonic-clonic activity. The presurgical evaluation was reviewed for all patients, including clinical history and

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examination, video-EEG monitoring data, brain MRI and PET scans, to identify the epileptogenic zone. A total of 17 patients (out of 6063 patients admitted to the EMU during that period of time) were identified with ITP as part of their seizure semiology. All patients identified were referred for presurgical evaluation of drug-resistant epilepsy. All had continuous video-EEG monitoring using standard scalp electrodes and eight patients were further monitored with intracranial EEG; four patients were monitored with StereoEEG (SEEG) and four with subdural grids. We reviewed the video recordings of all seizures with ITP. The direction of the ITP was determined as either clockwise or anticlockwise according to direction of movement of the body along the longitudinal axis of the body while looking from the foot end of the bed. A clockwise turn would be to the left and a counterclockwise turn to the right.

We reviewed intracranial EEG monitoring results where applicable, epilepsy surgery approach, and surgical outcome at last follow up.

3. Results

Among 17 patients identified, one was excluded due to inconclusive EEG localization. Sixteen patients (0.2%, out of 6063 patients monitored during this period) were included in the final analysis with twelve men and four women (Table 1). The average age was 32.5 years (range 18–50). An average number of eleven seizures (range 3–30) were recorded (Table 1). ITP was consistently seen in at least one focal impaired awareness seizure of all patients. Seizures with ITP started predominantly during sleep in 14 out of 16 patients; two patients had seizure onset during waking.

The ictal onset on scalp EEG was left temporal in five patients, right temporal in three (one of these patients was later found to have a right orbitofrontal focus with SEEG), left frontal in three, right frontal in five

Ten patients had ITP as part of hypermotor seizure semiology; ictal onset zone localization in scalp EEG in these patients was temporal lobe in five patients, probable frontal lobe in five patients.

The direction of ITP was uni-directional in 12 patients (2 clockwise; contralateral to ictal focus in one patient, 10 anticlockwise; contralateral to ictal focus in 5 patients and ipsilateral to the focus in 5 patients) and bi-directional in different seizures in four patients. Among these patients with bidirectional ITP, one patient had two clockwise and one anticlockwise seizure, his ictal onset was in the right temporal region for all these seizures. The second patient had one clockwise and one anticlockwise seizure with ictal onset in scalp EEG in the left temporal region that was further confirmed with subdural grids to be in left mesial temporal region. The third patient had three clockwise and two anticlockwise seizures with right temporal ictal onset on scalp EEG;

Table 1
Demographics and seizure semiology.

Patient	Gender	Age	Duration of epilepsy in years	Number of seizures recorded	Number of seizures with ITP
1	M	28	4	3	3
2	M	47	41	5	4
3	M	34	31	8	2
4	M	50	2	7	1
5	F	45	42	14	1
6	M	18	5	7	5
7	M	22	5	6	1
8	F	29	19	9	1
9	F	25	13	6	2
10	M	36	27	11	8
11	M	35	33.5	10	2
12	M	38	36.5	14	1
13	F	39	39	10	1
14	M	29	12	30	6
15	M	23	19	7	1
16	M	23	11	28	1

Table 2
MRI, Seizure localization and surgical outcome.

Patient	MRI abnormality	Localization on scalp EEG	Intracranial EEG	Intracranial localization	Direction of turning prone	Surgery	Surgical Outcome
1	Normal	Right temporal			2 clockwise, 1 anticlockwise		
2	Bifrontocentral encephalomalacia	Left frontal			4 anticlockwise		
3	Left occipitotemporoparietal encephalomalacia	Left temporal	SDG	Left hippocampus	1 clockwise, 1 anticlockwise	Left temporal lobectomy	IA
4	Left hippocampal sclerosis	Left temporal			1 anticlockwise		
5	Right frontal cortical dysplasia	Right temporal/ frontal	SEEG	Right inferior frontal	1 anticlockwise	RNS implanted	III
6	Normal	Right temporal	SEEG	Right orbitofrontal	3 clockwise, 2 anticlockwise	Right orbitofrontal resection	IA
7	Right cingulate cavernoma	Right midfrontocentral region			1 anticlockwise	Right cingulate resection	IA
8	Normal	Left frontal probable	SDG	Left cingulate	1 anticlockwise	Left cingulate resection	III
9	Normal	Right frontal probable	SDG	Right cingulate & SMA	2 anticlockwise	Right cingulate resection	IA
10	Right hippocampal sclerosis	Right temporal	SEEG	Right hippocampal	8 anticlockwise	Right SelAH	IA
11	Normal	Right frontotemporal convexity	SEEG	Bilateral hippocampus	2 anticlockwise	RNS implanted	III
12	Normal	Bifrontal/left frontal			1 anticlockwise		
13	Not Done	Right frontal probable			1 clockwise		
14	Normal	Right frontal	SDG	Right frontal	4 anticlockwise, 2 clockwise	RNS implanted	III
15	Left temporal cortical dysplasia	Left temporal			1 clockwise		
16	Normal	Possible left temporal			1 anticlockwise		

SDG: Subdural grid; SEEG: Stereo EEG; RNS: Responsive Neurostimulation; SMA: supplementary motor area; SelAH" selective amygdalohippocampectomy.

however SEEG demonstrated right orbitofrontal ictal onset. The fourth patient had two clockwise and four anticlockwise seizures with ictal onset in the right frontal convexity that was further confirmed with subdural grids.

MRI data were available in fifteen out of sixteen patients, as one patient did not have an MRI (Table 2). Seven patients had MRI abnormalities: mesial temporal sclerosis (MTS) in two patients, cavernous angioma in one patient, cortical dysplasia in two patients and encephalomalacia in two patients.

Nine patients underwent surgery (Table 2). Three patients had cingulate resection, one patient had temporal lobectomy, one had orbitofrontal gyri resection, and one patient had right selective amygdalohippocampectomy. Five patients had Engel class I outcome while one with left cingulate resection had Engel class III outcome. Three patients had a responsive neurostimulator (RNS) implanted, two in the right frontal convexity and one in bilateral hippocampal regions. All three patients had Engel class III outcome. Four patients were happy with their seizure control and refused surgery. Two patients were lost to follow up. One patient was deemed to be multifocal and not a surgical candidate

4. Discussion

Our study demonstrates that turning prone during seizure is not specific for frontal lobe epilepsy and can be seen in patients with temporal lobe ictal origin. In addition, the direction of turning is not lateralizing, and can be ipsilateral or contralateral to the ictal onset zone, and can even be in opposite directions in the same patient with unilateral ictal onsets. Thus ITP cannot be used as a localizing or lateralizing sign.

Ictal body turning of 90 ° or more along the horizontal body axis has been suggested as a distinct feature associated with mesial frontal lobe epilepsy [2]. Our study contradicts these findings and shows that ITP may also be seen in non-medial frontal lobe epilepsies. Saygi et al reported ITP not associated with versive head turns in five of their eleven patients and found it to be a useful ictal sign to differentiate frontal lobe focal seizures from PNES. They also excluded secondary generalized seizures [1]. However, their study was different from our study because they only compared FLE and PNES. Since they were not concerned with lateralization, they did not describe the direction of turning. Rheims and colleagues studied a cohort of patients with hypermotor seizures and found that Ictal body turning of 90 ° or more along the horizontal axis while lying down was associated with ictal onset in mesial premotor region. Their study differs from our study in that they did not exclude seizures that secondarily generalized, moreover they only compared hypermotor seizures with each other [8].

Gyratory, rotatory, circling and volvular seizures while standing have been previously used synonymously referring to rotation during seizure activity [9–11]. Seizures that have body rotation as a manifestation have been observed in generalized [12,13] as well as focal epilepsies [10,14,15]. In most published reports the circling was in transition to generalized tonic-clonic activity and may be related to versive head turning. In these instances it is distinct from the turning described in the current manuscript, which excludes versive head turning.

Seizures in which ITP is associated with versive head turning (usually in transition to bilateral tonic-clonic activity) usually involve the frontal eye field in Brodmann area 8, anterior to the precentral gyrus, which regulates voluntary saccadic eye and secondary head movement, while the ITP without versive head turning likely involves the basal ganglia and not the frontal eye field [16]. Prior studies in humans [17,18] and in experimental animals [19] have showed that basal ganglia may play a central role in rotational seizures [10,17,18] and spread of ictal discharge to basal ganglia results in rotational behavior. We postulate that bi-directional ITP in a given patient is likely due to different chronological stimulation of basal ganglia components.

Our study also showed that direction of ITP can be bi-directional in a substantial portion (4 out of 16 patients, 25%): some seizures in a given patient may have ITP with clockwise rotation while in other seizures in the same patient ITP may be anticlockwise. Shukla et al also described a cohort of 13 patients with turning of the whole body, with one patient turning to the left (anticlockwise) in one seizure and right (clockwise) in another seizure [20]. Their patient with bidirectional ITP had a left temporal epileptogenic lesion [20]. Usui et al described a cohort of patients with occipital lobe epilepsy and one out of their 13 patients had bidirectional versive movements. In two of the four seizures, versive movements were directed towards the side contralateral to the resection, whereas movements were ipsilateral in the remaining two seizures [21]. Interestingly one of our patients with bidirectional ITP had a right temporal lobe ictal focus, one a right orbitofrontal ictal onset, one a left temporal focus and one a right frontal focus. Out of these four patients, two underwent resective epilepsy surgery with Engel 1 outcome. One had RNS implanted in bilateral hippocampi with an Engel III outcome. Thus, our study suggests that bidirectional ITP is not an exclusion for epilepsy surgery.

Sixty two percent of patients in our cohort had hypermotor as well as ITP as a part of their seizure semiology. Hypermotor seizures consist of complex movements involving proximal segments of the limbs and trunk that appear violent [22]. Traditionally, hypermotor semiology has been thought to be associated with frontal lobe seizure origin [23]. However, recent reports have demonstrated an association of hypermotor seizures with other locations as well, such as temporal, insular, occipital and parietal lobes [21,24–26]. In Montavont study the frontal depth electrodes did not show ictal discharge except slow waves were observed during hypermotor seizures. Our study provides further support to the notion that hypermotor seizures and ITP may have extra frontal localization.

The limitations of our study include its retrospective nature. Second, our EEG data is based predominantly on surface EEG, which may have poor localizing value especially in frontal lobe epilepsy and conclusions based on scalp EEG only be misleading. However, surface EEG data was reviewed by two epilepsy specialists and only patients who had conclusive ictal changes at onset were included. Moreover, eight of our patients also had intracranial EEG monitoring to improve the localization. Third, seizure freedom after surgical resection represents the gold standard for localization of semiological signs. Surgical outcome was available in eight out of sixteen patients (50%) with five of these patients having Engel I outcome. Fourth, ITP during a seizure can represent a spread pattern. To address this, we excluded patients who had ITP as a late feature during their seizure or if seizures were secondary generalized. Fifth, our sample size is relatively small. We reviewed the EMU database for 12 years and we attribute small sample size to relatively rare presentation of this sign. The main implication of our study is that ITP is not a localizing or lateralizing semiological sign.

In summary, our study provides important data regarding ITP which is a rare and poorly understood ictal phenomenon. Our study confirmed findings of other investigators like Leung et al that ITP can be seen with various localization of the epileptogenic zone including mesial frontal, lateral frontal, parietal, occipital or temporal lobe. ITP does not have a consistent localizing or lateralizing value, and may be bi-directional from a single epileptogenic focus.

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