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Genital automatisms: Reappraisal of a remarkable but ignored symptom of focal seizures

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ABSTRACT

Objectives: Genital automatisms (GAs) are uncommon clinical phenomena of focal seizures. They are defined as repeated fondling, grabbing, or scratching of the genitals. The aim of this study was to determine the lateralizing and localizing value and associated clinical characteristics of GAs.
Methods: Three hundred thirteen consecutive patients with drug-resistant seizures who were referred to our tertiary center for presurgical evaluation between 2009 and 2016 were investigated. The incidence of specific kinds of behavior, clinical semiology, associated symptoms/signs with corresponding ictal electroencephalography (EEG) findings, and their potential role in seizure localization and lateralization were evaluated.
Results: Fifteen (4.8%) of 313 patients had GAs. Genital automatisms were identified in 19 (16.4%) of a total 116 seizures. Genital automatisms were observed to occur more often in men than in women (M/F: 10/5). Nine of fifteen patients (60%) had temporal lobe epilepsy (right/left: 4/5) and three (20%) had frontal lobe epilepsy (right/left: 1/2), whereas the remaining two patients could not be classified. One patient was diagnosed as having Rasmussen encephalitis. Genital automatisms were ipsilateral to epileptic focus in 12 patients and contralateral in only one patient according to ictal–interictal EEG and neuroimaging findings. Epileptic focus could not be lateralized in the last 2 patients. Genital automatisms were associated with unilateral hand automatisms such as postictal nose wiping or manual automatisms in 13 (86.7%) of 15 and contralateral dystonia was seen in 6 patients. All patients had amnesia of the performance of GAs.
Conclusion: Genital automatisms are more frequent in seizures originating from the temporal lobe, and they can also be seen in frontal lobe seizures. Genital automatisms seem to have a high lateralizing value to the ipsilateral hemisphere and are mostly concordant with other unilateral hand automatisms. Men exhibit GAs more often than women.

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1. Introduction

Ictal clinical lateralizing signs are particularly beneficial in presurgical investigation of refractory epilepsy. Some of these frequent or rare semiologic phenomena have well-known high lateralizing and localizing value reaching 90–100%, whereas some others seemed less reliable [1–4]. An exhaustive body of literature has evaluated many of these semiologic features, whereas GAs remain largely underinvestigated. Their frequency has been reported as 3–11.4%, which suggests that this phenomenon is not rare but underestimated. Genital automatisms occurred relatively frequently in temporal lobe seizures (16%) and relatively rarely (4%) in frontal lobe epilepsy (FLE) [5]. The lateralizing value of GA is controversial.

Genital automatisms defined as repeated fondling, grabbing, or scratching of the genitals are detected in focal seizures [5]. They must be differentiated from other sexual and orgasmic auras, genital sensory phenomena, and hypermotor sexual automatisms. The localizing value for these phenomena is debatable. It was reported that subtle genital automatisms such as fondling and genital grabbing were more typical of temporal lobe seizures, whereas hypermotor seizures such as pelvic or truncal movements were prevalent in FLE [6].

We aimed to evaluate the lateralization and localization value of GAs and associated clinical characteristics to draw attention to these underestimated epileptic phenomenon as a valuable semiologic characteristic.

2. Material and methods

2.1. Patient selection

We retrospectively studied 313 consecutive patients with medically refractory seizures who were admitted to the epilepsy monitoring unit at the Epilepsy Center, Istanbul Faculty of Medicine, Istanbul University,

Abbreviations: EEG, electroencephalography; FLE, frontal lobe epilepsy; GAs, genital automatisms; LOC, loss of consciousness; MRI, magnetic resonance imaging; PET, positron-emission tomography; SUDEP, sudden unexpected death in epilepsy; TLE, temporal lobe epilepsy; VEM, video-EEG monitoring.

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between 2009 and 2016, for the presence of GAs. All patients underwent long-term video-EEG monitoring (VEM) for more than 24 h with scalp electrodes, in addition to their detailed presurgical evaluation. Clinical and electrophysiologic seizures showing GAs were included and evaluated with a standard protocol [7].

2.2. Data collection tools

The video-electroencephalography (EEG) recording technique, performed using scalp electrodes (International 10–20 System), was in accordance with international guidelines developed by the American Electroencephalographic Society for video-EEG monitoring in patients with epilepsy [8]. Scalp EEG analysis was performed using bipolar longitudinal-transverse and referential montages by experienced clinical neurophysiologists. The EEG recordings of all patients were examined to evaluate the subsistence of background abnormalities, both nonepileptiform and epileptiform interictal activity, ictal discharges, and postictal slowing. Additionally, we evaluated interictal scalp EEG, ictal semiology, and neuro-imaging findings. High-resolution 3 T MRI was performed according to a standard protocol, and interictal PET and formal neuropsychological evaluations were performed. We evaluated invasive ictal EEG in one patient (N7). Epileptic focus definition was based on accordant findings in these investigations. Our main indicator was the scalp EEG ictal signs to determine the seizure onset zone. The other signs were accepted as supplementary methods. If we could not lateralize the seizure onset zone with the main or supplementary methods, we described this as "nonlateralized". One patient (N7) who had two seizures with GAs showed switch of lateralization phenomenon. If the scalp EEG ictal signs lateralization switched to the contralateral hemisphere, we accepted this phenomena as "switch of lateralization". Bilateral asynchrony was more than 1 Hz frequency difference between the two sides and the duration must be at least 10 s [9].

2.3. Genital automatisms

We defined GAs as stereotyped, repetitive manipulation of genitals for grabbing, scratching, or rubbing, for longer than 3 s [5]. We categorized GAs according to the time of occurrence: "early" period: first 10 s from the beginning of seizure; "late" period, the last 10 s; "intermediate" period, between these; and "postictal" period, after the end of seizure activity on EEG. All video-EEG records were reviewed by two examiners (NB, HOD) who were blinded to the history and seizure outcome in order to detect seizures with GAs, and to evaluate their electroclinical semiology. Consensus was established for all patients.

Genital sensations such as numbness, tingling, pain, and unpleasant feeling in the genitals are classified as somatosensory auras. Sexual auras are usually experienced as erotic pleasant feelings or thoughts [10]. None of our patients reported any of these sensations.

We used the Engel classification system for postoperative outcomes in patients who had epilepsy surgery [11,12]. Class I: free of disabling seizures, Class II: rare disabling seizures ("almost seizure-free"), Class III: worthwhile improvement, and Class IV: no worthwhile improvement.

All patients were informed, and written consent was obtained. The local ethics committee approved the study (2015-1029).

3. Results

We observed GAs in 15 (4.8%) of 313 patients. The mean age of patients was 32.3 ± 9.4 years (range, 15–50 years). The majority (10 men and 5 women; 66.7%) of patients who exhibited GAs were men.

The mean seizure onset age was 13.2 ± 10.5 years (range, 1–38 years). The mean seizure frequency for the last three months was 4.9 ± 4.19 (range, 1–15). One hundred sixteen seizures in total were recorded from these patients. We detected GAs in 19 of 116 seizures. Three patients had GAs in multiple seizures. The 10th patient had 3, and the 7th and 9th patients had 2 seizures with GAs; the remainder had only one (Table 1).

All GAs were subtle hand automatisms, none involving pelvic thrusting or hypermotor behavior. Of the 16 ictal GAs, 6 were seen in the early period, 7 in the late, and 3 in the intermediate periods. Genital automatisms were seen postictally in three seizures.

Nine patients had hippocampal sclerosis, 3 patients had frontal encephalomalacia, and 1 patient had Rasmussen encephalitis. Two patients had no lesions.

During VEM, all patients who performed GAs had focal seizures with LOC and were amnestic of the GAs. The patient with Rasmussen encephalitis had a secondarily generalized focal seizure.

No patients reported a history of a genital somatosensorial aura. None of the patients experienced any apparent autonomic symptoms such as orgasm during the seizures associated with GAs, and in male patients, no penile erection or ejaculation was observed.

According to EEG findings, ictal seizure onset was localized in the anterior temporal region in 8 patients. Genital automatisms were on the same side as the ictal onset in seven patients (87.5%). Only in one patient (N2) the ictal activity started in the right temporal region, and GA occurred on the contralateral (left) side. Ictal onset was in the left frontal lobe in one patient (N11) and in bilateral anterior regions in another patient (N1); it could not be localized in 5 patients with ictal scalp EEG. In four of these patients whose ictal EEGs were uninformative, ictal semiology, interictal EEG, and cranial MRI findings were concordant with the lateralization of GAs to the ipsilateral hemisphere (Fig. 1A-B). Two patients could not be lateralized using scalp EEG or other auxiliary methods (Fig. 2).

One patient (N7) who had two seizures with GAs showed switch of lateralization phenomenon ictal activity began in the left temporal region and spread to the right temporal region. Genital automatisms started in the left hand, then continued in the right hand with simultaneous ictal seizure activity (Fig. 3A-B). This patient was additionally evaluated with invasive EEG (bilateral deep hippocampal electrodes), because of bilateral ictal, interictal electrophysiological and radiological findings. Two seizures were recorded during invasive EEG, which showed seizure onset in the left anterior temporal region, ipsilateral to GAs (Fig. 4).

Genital automatisms were seen together with unilateral hand automatisms such as postictal nose wiping or manual automatisms in 13 (86.7%) of 15 patients accompanied by unilateral dystonia in 6 (42.8%) patients. Unilateral manual automatisms were ipsilateral and concordant with GAs in 11 (84.6%) patients, unilateral dystonias were 100% contralateral with GAs.

Four patients underwent to epilepsy surgery. Two had good outcomes (Engel's class 1) with a follow-up of ≥2 year. Two patients had surgery within the past 6 months (N7, N12) with no postoperative complications. Four patients declined surgery. One patient was evaluated recently and is awaiting surgery. One patient with right hippocampal sclerosis (N5) who was a candidate for surgery died before surgery in 2012 of probable SUDEP. This patient experienced 5–6 seizures per month despite multiple antiepileptic drugs. He had no cardiac disease or history of any other illness.

4. Discussion

Genital and sexual seizure manifestations represent underinvestigated and rare clinical phenomena during or after seizures and can be subdivided into (1) sexual auras, (2) genital auras, (3) sexual automatisms, and (4) GAs [13]. The frequency of GAs was reported as 3–11.4% [5]. Genital automatisms were detected in 4.8% of our patients. A possible role of the temporal lobe in sexual behavior was originally reported by Klüver and Bucy [14] who demonstrated that bilateral temporal lobectomy in monkeys produced hypersexuality. Previous studies that investigated the lateralization and localization value of

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