Genital Automatisms: A Video-EEG Study in Patients with Medically Refractory Seizures


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Summary: Purpose: Genital automatisms (GAs) are rare clinical phenomena during or after epileptic seizures. They are defined as repeated fondling, grabbing, or scratching of the genitals. The anatomic correlates of GAs have been discussed controversially. The aim of this investigation was to assess the localizing and lateralizing value of GAs.

Methods: The authors studied 207 consecutive patients with intractable seizures referred to a University Hospital for presurgical evaluation between 1998 and 2002: 135 had temporal lobe epilepsy (TLE); 23, frontal lobe epilepsy (FLE); 29, generalized epilepsies (GEs); and 20 had extratemporal or multifocal epilepsy.

Results: Twenty-three (11%) of 207 patients showed GAs in 42 (3%) of 1,299 seizures. GAs occurred significantly more often in men (17 of 93, 18%) than in women (six of 114, 5%; p = 0.0037). Twenty-one (16%) of 135 patients with TLE performed GAs, one (4%) of 23 with FLE and one (3%) of 29 with GE. GAs were associated with unilateral hand automatisms in 16 (70%) of 23 and with perictal urinary urge in five (22%) of 23. All patients had amnesia for the performance of GAs.

Conclusions: GAs appear in the ictal or postictal period with impaired consciousness. Men exhibit GAs significantly more often than do women. GAs do not localize or lateralize per se, but may localize seizure onset in the presence of perictal urinary urge or unilateral hand automatisms. They show a tendency to occur more often in TLE. Key Words: Genital automatisms—Sexual seizure symptoms—Video-EEG seizures—Presurgical epilepsy evaluation.

Genital automatisms (GAs), defined as repeated fondling, grabbing, or scratching of the genitals, are a rare symptom during or after epileptic seizures. They must be separated from other genital or sexual seizure manifestations (1), like sexual and orgasmic auras (2–7), genital sensory phenomena (7–11), and hypermotor sexual automatisms (3, 4,12, 13). The localizing value for these phenomena is discussed controversially. Whereas some authors suggested a temporal seizure onset for various types of sexual seizure phenomena (2–6, 14), others reported a seizure-onset zone in the frontal lobe (12,13). Pure GAs, as defined earlier, have been observed in 6% of patients with medically refractory complex partial seizures, and the authors suggested a temporal seizure-onset zone in their patients (15).

We wanted to clarify whether GAs are related to a specific brain region and add valuable information on localizing and lateralizing the seizure-onset zone or whether they are a nonspecific feature of epileptic seizures.

PATIENTS AND METHODS

We retrospectively studied 1,299 seizures in 207 consecutive patients: 93 men and 114 women, ranging in age from 17 to 75 years, with a mean of 39.4 ± 13 years. One hundred thirty-five patients had temporal lobe epilepsy (TLE); 23, frontal lobe epilepsy (FLE); 29, generalized epilepsies of different types (GEs); and 20, extratemporal or multifocal epilepsy. They were admitted to the epilepsy-monitoring unit at the Universitätsklinik Innsbruck, Austria, between February 1998 and October 2002 because of medically refractory seizures. All patients underwent long-term video-EEG monitoring with scalp and sphenoidal electrodes for an average of 5.4 days (range, 2–13 days). We defined GAs as stereotyped, repetitive manipulation of the genitals for ≥3 s: grabbing, scratching, or rubbing, and masturbating. They were called “ictal,” occurring before EEG seizure offset (16), or starting in the ictal and continuing in the postictal period. GAs occurring after the EEG seizure offset were called “postictal.” We defined the end of the postictal period as the clinical normalization of cognitive and motor function and the return to the habitual interictal EEG.
Two reviewers (J.D., G.W.) examined the seizures individually. The occurrence of GAs was determined by scoring a sign as present if it was reported by both reviewers. If the two reviewers disagreed, the video was reexamined by a third reviewer (E.T.). The overall interobserver agreement was excellent, with a kappa index of 0.97.

We identified 23 patients with GAs. They underwent a comprehensive presurgical evaluation including a detailed clinical examination, seizure history, and prolonged video-EEG monitoring. Patients were specifically asked for a genital aura or any libidinous psychosexual aura experienced during their habitual seizures. A high-resolution magnetic resonance imaging (MRI) according to a standard protocol was done, with interictal single-photon emission computed tomography (SPECT) or positron emission tomography (PET), formal neuropsychological evaluation, and a Wada test or functional MRI (fMRI) to lateralize language and memory dominance, when feasible. Seizure-focus definition was based on concordant findings in these investigations.

**RESULTS**

We observed GAs in 23 (17 men and six women) of 207 patients, representing 11% of all patients, and in 42 of 1,299 seizures, representing 3% of all recorded seizures. Mean age of patients was 42.7 ± 11.2 years (range, 1–64 years). The majority (74%) of patients who exhibited GAs were men (Fisher’s exact, p = 0.0037). Twenty-one patients had TLE, one had FLE, and one patient had GE. A trend for GAs to occur more often in seizures from the temporal lobe was noted, but this did not reach significance (χ² test, p = 0.0916).

Twenty-eight (67%) of the seizures associated with GAs were complex partial. In 21 of them, an aura was present; seven were complex partial from the onset. Thirteen (31%) seizures were secondarily generalized; seven of them started with an aura. One (2%) of 42 seizures was generalized.

All patients who performed GAs had amnesia for that activity. None of the patients reported a history of a genital somatosensory aura, but one had a sexual orgasmic aura (“tingling feeling through the body like an orgasm”). None of the patients apparently experienced orgasm during the seizures associated with GAs, and in male patients, no penile erection or ejaculation was observed.

GAs started ictally in 12 patients; all of them had TLE. In 11 patients, GAs were performed exclusively postictally. Nine of them had TLE; one patient, FLE; and one patient, GE (χ² test, p = 0.3027). Of 42 seizures, 20 were associated with ictal GAs, and 22, with postictal GAs. The surface EEG at the onset of GAs showed bilateral temporal seizure pattern or bilateral slowing in 34 (81%) of 42 seizures. The surface EEG showed unilateral temporal seizure pattern or unilateral slowing in eight (19%) of 42 seizures at the onset of GAs.

Mean duration of GAs was 128 ± 240 s (median, 51 s; range, 3–953 s) in men and 37 ± 18 s (median, 40 s; range, 17–65 s) in women. In 16 (70%) patients, the GAs were associated with unilateral hand automatisms of the same hand used for performing GAs. In five (four men, one woman; 22%) patients, GAs were associated with pericentral urinary urge (Table 1).

Thirteen patients proceeded subsequently to resective surgery. Nine of them had an excellent outcome (Wieser class 1) with a follow-up of ≥1 year; one patient experienced a worthwhile reduction in frequency and intensity of his seizures (Wieser class 4) (17). In two patients, seizure frequency did not change remarkably (Wieser class 5), and in one patient, postoperative follow-up was too short for meaningful analysis.

**DISCUSSION**

We report 23 patients, who repeatedly manipulated their genitals during or immediately after seizures. GAs occurred in seizures originating from the temporal lobe, frontal lobe, as well as in generalized seizures. Men exhibited GAs significantly more often than did women. GAs were associated with unilateral hand automatisms in 70% and with pericentral urinary urge in 22%.

In patients with epilepsy, the localizing value of ictal or pericentral sexual behavior is discussed controversially. A report on four patients with sexual automatisms, who were studied with depth electrodes, found a seizure-onset zone in the frontal lobe portion of the limbic system (12). The authors did not separate hypermotoric sexual automatisms from discrete GAs, and only two patients remained seizure free after frontal resections. One patient with discrete GAs was not seizure free, and the other one was not operated on. Another report on 10 patients with hypermotoric activities like pelvic thrusting, kicking, and rocking, as well as discrete manipulations of the genitals, demonstrated a seizure onset in medial or orbital frontal region with depth electrodes (13).

In contrast to these findings, a later report on GAs, with careful analysis of the semiology of sexual behavior in patients who were rendered seizure free after limited temporal resection, suggests that repeated fondling or grabbing of genitals, during or immediately after seizures, is more common with temporal lobe seizure onset (15). The authors distinguished GAs from hypermotoric sexual automatisms, like pelvic or truncal thrusting, eventually combined with repeated grabbing, which are more common in seizures originating in the frontal lobe (12,13). However, in their report, only five of 74 patients with TLE had GAs, whereas none of the 16 patients with FLE had GAs. The sample size may have been too small to detect any patients with extratemporal seizure onset and GAs.
In our study, the sample size and the number of patients with GAs was larger (n = 23). We found one patient with frontal lobe seizure onset and one with generalized seizure onset, who revealed subtle repeated grabbing of the genitalia as defined earlier. Although a trend was noted for GAs to occur more often in patients with TLE, this was not significant. One could speculate that the GAs found in our patient with presumed seizure onset in the frontal lobe could be falsely localized because of seizure spread from the temporal into the frontal lobe. We cannot exclude this, because we did not operate on the patient, and he refused depth electrode studies for further localization.

Although most reports describe GAs in patients with focal seizures, they also have been described in patients with GE (18). Our patient with generalized seizure onset was admitted for resective surgery because of some focal EEG findings, as sometimes found in patients with GEs. This fact may imply some kind of "secondary temporalization" (19). A definite localization with depth electrodes in this patient was not applicable.

The mechanisms for GAs are far from clear. A possible role of temporal lobe structures in human sexual behavior was suggested by Klüver and Bucy (20) in 1939. They analyzed monkeys (after bilateral temporal lobectomy) that exhibited indiscriminate hypersexuality, excessive oral tendencies, excessive attentiveness to visual stimuli, and psychic blindness, together with masturbatory activity. Later reports of humans with bitemporal damage after severe brain trauma or encephalitis confirmed these observations (21,22).

The similarity of epileptic GAs to the behavior found in human Klüver–Bucy syndrome suggests a transient bitemporal dysfunction due to ictal epileptic activity or postictal dysfunction. The seizure onset may be generated in the temporal lobe or spread to the temporal lobes from frontal or parietal neocortex. A case report of a patient with recurrent postictal Klüver–Bucy syndrome, which occurred transiently after seizures in a woman who had occasional seizures after unilateral temporal lobectomy, strongly supports this hypothesis (23). The EEG at onset of GAs in our patients showed a bitemporal seizure activity or bilateral slowing in 34 (81%) seizures, whereas a unitemporal seizure activity or unilateral slowing was recorded only in eight (19%) seizures. This also supports the hypothesis of transient bitemporal dysfunction leading to GAs.

Alternatively, GAs may occur as a nonspecific reaction to an internal stimulus, like periictal urinary urge. Previous studies found an incidence of periictal urge to void in 0.4 to 2.2 in a comparable patient population (24,25). We found GAs associated with periictal urinary urge in five patients (four men, one woman; 22%); four of them had a seizure onset in the nondominant temporal lobe. Thus we suggest that some patients, mostly men, exhibiting GAs have a periictal urinary urge to void and therefore grab
their genitalia repeatedly. The majority of these patients had a seizure onset in the nondominant hemisphere. However, we cannot conclude from our data that GAs have any lateralizing value per se. They tend to lateralize only if they were associated with unilateral hand automatisms or with perictal urinary urge.

We consider GAs during epileptic seizures to be a non-specific behavioral pattern in the presence of bitemporal dysfunction or a response to internal stimuli. GAs may localize seizure onset to the ipsilateral temporal lobe only when they are associated with unilateral hand automatisms or to the nondominant temporal lobe when associated with perictal urinary urge.

REFERENCES