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COMMENTARY

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Gyratory seizures as a presentation of temporal encephalocele

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Abstract

Gyratory seizures (GS) are rare and have been reported in focal (frontal and temporal) as well as generalized epilepsies. The exact neuroanatomical substrate of gyration during a seizure is not clearly understood, but is thought to be due to the involvement of the basal ganglia. The direction of gyration occurring without head version is thought to have ipsilateral cerebral origins, while gyrational seizures preceded by forced head version are suggestive of seizure onset contralateral to the direction of gyration or the presence or absence of forced head version is thought to have important implications for lateralization of seizure origins from the direction of gyration. This case describes gyratory seizures in a young boy with a temporal encephalocele. Report of MRI brain initially indicated no abnormalities, but careful review revealed a left temporal encephalocele. PET scan showed left temporal hypometabolism. The patient underwent a left anterior temporal resection with amygdalohippocampectomy (ATL + AH) and is seizure-free for 18 months. Temporal encephalocele is the most commonly iatrogenic but may also be spontaneous, posttraumatic, or in relation to chronic otitis media. This case suggests that gyratory seizures may be a unique presentation of temporal encephalocele and this possibility warrants investigation in patients with medically refractory epilepsy.

KEYWORDS

encephalocele, gyratory seizures, temporal lobe epilepsy

1 | INTRODUCTION

Gyratory seizures (GS) are a rare occurrence, but have been reported in focal seizures of frontal and temporal origins, as well as generalized epilepsies.[1–3] GS are characterized by rotation around the body axis in a circular or spiral manner, typically by at least 180 degrees or 360 degrees.[2] The term gyratory has also been used to refer to volvular, circling, or rotatory seizures and has been associated with hamartomas, structural malformations, hippocampal sclerosis, thalamic gliomas, and nonlesional cases as well.[1–5] Very few cases, however, have been reported in the context of epilepsy with a temporal encephalocele. The current case describes this unique seizure semiology in a young boy with this structural defect.

2 | CASE

A 16-year-old boy, SB, presented with recurrent seizures since 12 years of age. He characterized his aura as consisting of abnormal thoughts of something "indescribable." Seizure

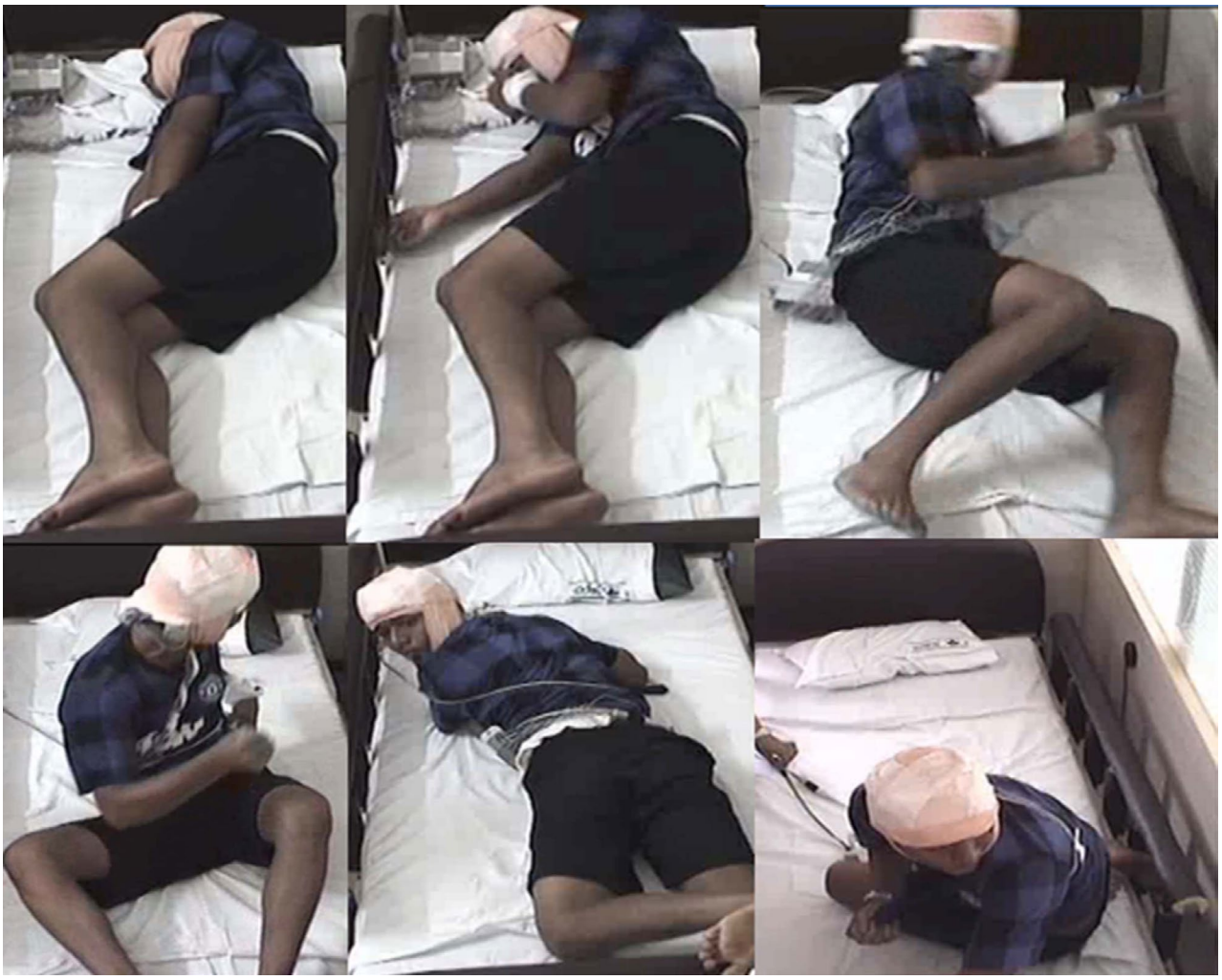

FIGURE 1 Patient lying in right lateral position followed by holding nose with left hand, followed by right hand dystonic posturing subsequently gyration to left with cables wrapped around him, later sitting in bed with looking toward left with right hand postured

semiology was reported to involve gyration to the left side five to six times with dystonic posturing of his right hand and pouting for 1-2 minutes. The patient also reported amnesia for these events. SB would experience gyratory movements both in standing as well as in the supine position and had never experienced a generalized seizure. Seizure frequency was reported to be 1 per week, with the occurrence of clustering.

Video electroencephalography (V-EEG) recorded a total of four stereotyped gyratory seizures. All seizures were characterized by gyration to the left, initially with right-handed nose holding followed by dystonic posturing of the right hand, confusion, and then continuous gyration to left, five to six times with a prominent stare and pout. Each seizure lasted 90-120 seconds, without postictal aphasia, although amnesia for the event was present (Figure 1). Ictal EEG showed left anterior temporal polymorphic theta rhythm and then becoming a spiky theta rhythm. Ten seconds later, this was found to spread to the central chain and right temporal region, evolving into theta delta rhythm with artifacts. Interictal record showed left temporal spike and wave discharges (Figure 2). MRI brain was initially reported as being normal, but on multiple reviews by different clinicians, a left temporal encephalocele was identified. PET scan showed left temporal hypometabolism (Figure 3).

Given concurrence of findings on structural and functional neuroimaging, the patient underwent resection of the left temporal encephalocele with anterior temporal resection and amygdalohippocampectomy (ATL +AH). Postoperatively, SB has been seizure-free for 18 months and is no longer on antiepileptic medication. The unique feature of this case is the presence of gyratory seizures as a manifestation of a temporal encephalocele.

3 | DISCUSSION

Gyratory seizures are rare, and very few studies have described associated V-EEG findings in detail.[1] Earlier studies reported generalized etiology to be more common

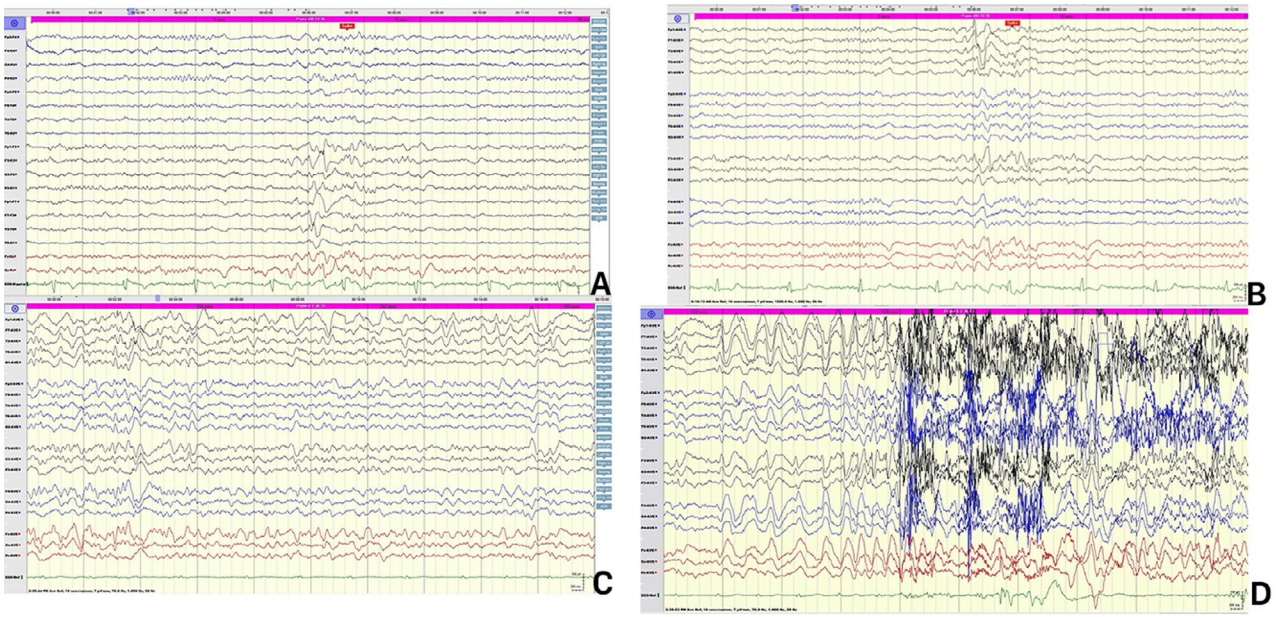

FIGURE 2 A and B, Interictal EEG showing left temporal spikes. C and D, Ictal recording showing left temporal polymorphic theta rhythm followed by spiky theta rhythm subsequently associated with muscle artifact

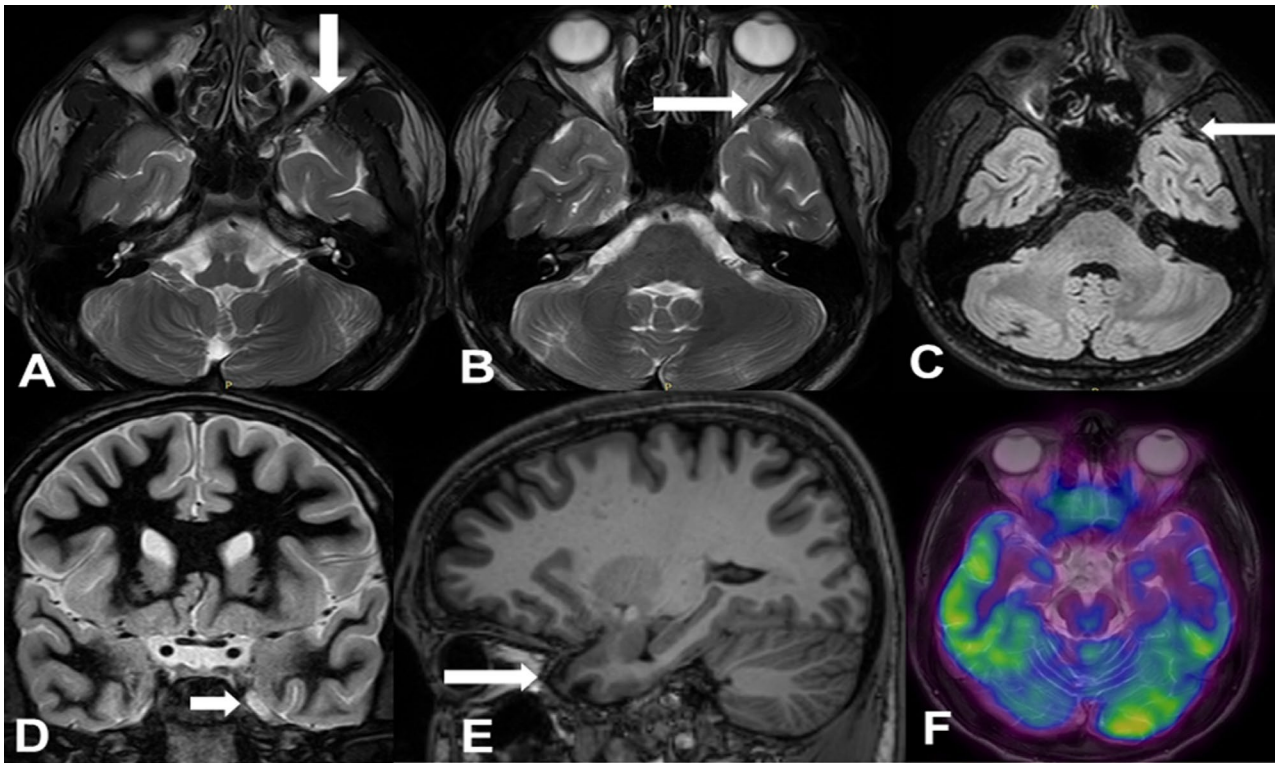

FIGURE 3 A and B, Axial T2 W; C, Axial FLAIR; D, Coronal; E, Sagittal MRI showing left temporal encephalocele (arrow); F, MRI PET fusion image showing left temporal hypometabolism

but this was based on history alone. In contrast, a study by Dobesberger et al reported all of their patients to experience focal seizures—eight with frontal and four with temporal onset (three with hippocampal sclerosis and one with hippocampal atrophy).[1] One patient had closed lip schizencephaly, and one had a cingulate gyrus hamartoma. Gyratory seizures were also described in the presence and absence of forced head version,[1] with this feature having lateralizing value. Specifically, the direction of gyration without head version is thought to be ipsilateral to the side

of seizure onset, while the direction of gyration in a seizure preceded by forced head version is presumed to be contralateral to the side of seizure onset.

In the case of SB, the seizure involved gyration to the left without forced head version, ipsilateral to the locus of the temporal encephalocele as would be predicted. In addition, dystonic posturing of the right hand also suggests left-sided lateralization. The exact mechanism of gyration during a seizure is not clearly understood, but is thought to be due to the involvement of the basal ganglia. In the study by Veruceil et al, volvular seizures originated from the anterior and mesial part of the right temporal lobe with single photon emission computed tomography (SPECT) revealing rapid ipsilateral orbito-cingular involvement.[6] SB's observed semiology of left-sided gyration with dystonic posturing of right hand, accompanied by an ictal EEG showing left temporal onset, was suggestive of a seizure originating from the left temporal lobe with spread to the basal ganglia resulting in dystonia.

Very few studies have reported a temporal encephalocele resulting in epilepsy.[7] The largest series of temporal encephalocele consists of 23 patients and was reported by Saavalainen et al. Standard MRI examinations in newly diagnosed epilepsy estimated the frequency of a temporal encephalocele to be 0.3%, with a slightly higher frequency of 1.9% in patients with drug-resistant epilepsy. None of the patients in their series had gyratory seizures, and 12 underwent surgery with a good outcome.

It is worth noting that SB was initially thought to be MRI-negative with PET positivity (left temporal hypometabolism), and the temporal encephalocele was only identified on meticulous, repeated review of MRI brain. It then follows that in all cases of MRI-negative temporal lobe epilepsy, a careful review for an encephalocele may prove to be useful. Most commonly iatrogenic in origin, temporal encephaloceles can also be observed post-trauma or in relation to chronic otitis media.[7,8] There was however no history of trauma or otitis media in SB.

Surgery in temporal encephalocele can involve focal resection of encephalocele or ATL+AH. The literature reports good outcomes in temporal lobe epilepsy with encephalocele,[7,8] and results of case SB are consistent with this finding. Thus, on encountering gyratory seizures of temporal onset, the possibility of a temporal encephalocele should be considered.

4 | CONCLUSION

The current case suggests that gyratory seizures can be a unique presentation of temporal encephalocele. In patients with medically refractory epilepsy, careful evaluation of MRI for temporal encephalocele is warranted as it is associated with a good surgical outcome.

DISCLOSURE

Neither of the authors has any conflict of interest to disclose. We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

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