Epilepsy & Behavior Case Reports 6 (2016) 49–51

Contents lists available at ScienceDirect

Epilepsy & Behavior Case Reports

journal homepage: www.elsevier.com/locate/ebcr

Case Report

'Tickling' seizures originating in the left frontoparietal region

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ARTICLE INFO

Article history:
Received 22 June 2016
Received in revised form 7 July 2016
Accepted 9 July 2016
Available online 29 July 2016

Keywords:
Sensory seizures
PNES
Frontal epilepsy
Parietal epilepsy
Tickling
Behavioral stereotypy

ABSTRACT

We report a 10-year-old boy with mild developmental delay and epilepsy with new events of right back tickling and emotional upset. These initially appeared behavioral, causing postulation of habit behaviors or psychogenic nonepileptic seizures. Several ictal and interictal EEGs were unrevealing. Continuous EEG revealed only poorly localized frontal ictal activity. Given that his clinical symptoms suggested a parietal localization, double-density EEG electrodes were placed to better localize the epileptogenic and symptomatogenic zones. These revealed evolution of left greater than right frontoparietal discharges consistent with seizures at the time of the attacks. Medical management has significantly reduced the patient's seizures.

1. Introduction

Parsing true epilepsy from behavioral stereotypy and psychogenic nonepileptic seizures (PNES) is extremely important. The prevalence of epilepsy is 5 to 10 per 1000, and the estimated prevalence of PNES is 2 to 33 per 100,000, making them both significant diseases [1]. In children, consideration of behavioral stereotypies is also very important, as these are extremely common, by some estimates occurring in up to one-third of all children [2]. While behavioral stereotypies are less commonly confused with epilepsy than with PNES, they are frequently coexistent with both and are more common in patients with developmental delay [3]. (See Fig. 1.)

Misdiagnosing patients with true epilepsy as suffering from PNES can be catastrophic. Not only do their seizures go untreated until the correct diagnosis is made – putting the patient at increased risk of all the problems associated with untreated epilepsy – but it also causes significant problems when the correct diagnosis is made. These patients are often distrustful of the medical community and are less compliant with necessary medications or other treatments for their seizures and question the validity of their new diagnosis. Equally important is diagnosing PNES accurately. The sooner PNES is diagnosed the better the cure rate, which is particularly true in children, who have a more favorable outcome than adults [4]. While behavioral stereotypies can be upsetting to families, their lack of treatment does not cause harm. Misidentification of these as nonepileptic behaviors when they in fact are seizures is disastrous, as the patient's epilepsy then goes untreated.

Parietal lobe seizures are relatively rare compared with frontal or temporal lobe seizures and can have many different semiologies. This can make the identification of clinical features more difficult and their confusion with PNES or behavioral stereotypies more likely. Treatment of the abnormal behaviors with therapy and psychotropic medications can aid in distinguishing PNES and behavioral stereotypies from true epilepsy.

Our case describes a patient with an interesting clinical presentation of sensory seizures as well as a dramatic behavioral overlay, which presented as a new seizure semiology for him. It illustrates the importance of further investigation for possible psychogenic spells and how double-density EEG electrodes may help to clarify a suspected localization.

2. Methods

Routine EEGs as well as the initial continuous video-EEG (cEEG) recording used 19 MRI-compatible electrodes plus 2 reference and 2 EKG leads. These were placed using the standard 10–20 International system of electrode placement. Twenty-one hours into continuous EEG (cEEG) monitoring, double-density electrodes were placed over the bilateral frontal and parietal head regions. The additional electrodes added included the following: FC3, FC4, CP3, CP4, FCz, and CPz, which were placed using the 10–10 system.

Abbreviations: PNES, psychogenic nonepileptic seizures; cEEG, continuous electroencephalogram; MEG, magnetoencephalography; SISCOM, subtracted ictal spect coregistered to MRI brain; SSRI, selective serotonin reuptake inhibitor; CBT, cognitive–behavioral therapy.

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http://dx.doi.org/10.1016/j.ebcr.2016.07.002

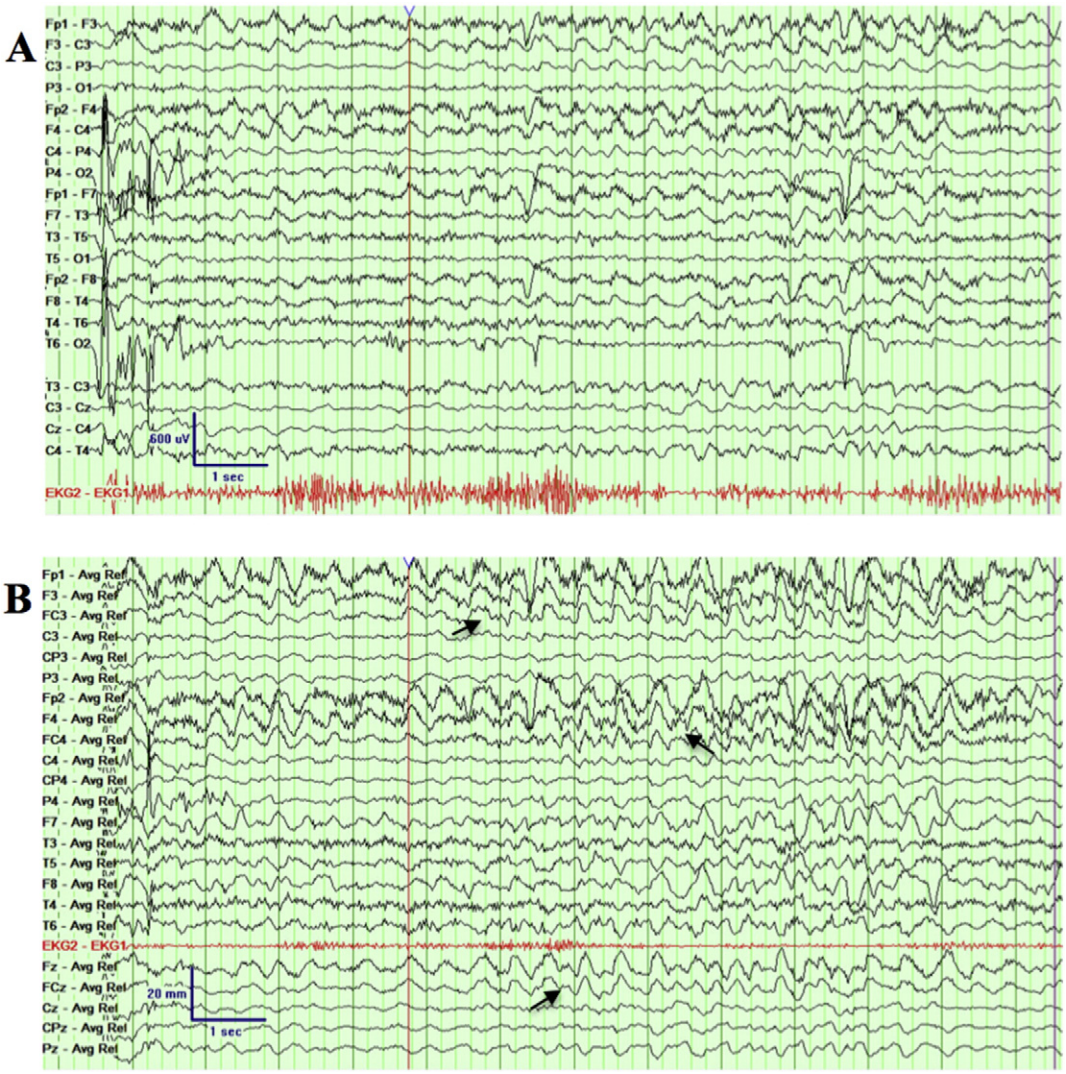

Fig. 1. Epileptiform activity during a seizure (amplitude: 30 μV/mm, filter: 30 Hz): A) bipolar montage, B) average reference montage with double-density electrodes over the frontoparietal region. Rhythmic 1- to 2-Hz activity is seen over the frontoparietal region, which is more clearly seen in FC4, FC3, and Fz with the double-density electrodes. Arrows point out the epileptiform activity.

3. Case study

A 10-year-old male with history of hydrops fetalis, developmental delay, learning impairment, and long-standing focal epilepsy, which had been solely subclinical since the age of two, presented with complaints of recurrent and painful 'tickles' in his right lower back.

The patient's epilepsy is thought to be due to hydrops fetalis. He was born at 35 weeks by emergent cesarean section due to fetal heart decelerations and maternal hypotension. He required resuscitation twice after birth, having APGARS of 1 at 1 min, 2 at 5 min, 1 at 10 min, 1 at 15 min, 5 at 20 min, 6 at 25 min, and 7 at 30 min. Head ultrasound shortly after birth was normal. The electroencephalogram (EEG) at that time was abnormal because of reduced reactivity and discontinuity — but showed no epileptiform abnormalities. Initially, his development was only slightly delayed. However, his cognitive skills were noted to be more significantly delayed when he started school and have continued to lag. He was diagnosed with learning disability.

He underwent formal neuropsychological evaluation at the age of eight. On the Wechsler Intelligence Scale for Children—Fourth Edition (WISC-IV), he performed in the extremely low range (full-scale IQ = 57) when compared with other students his age, placing him at the 0.2 percentile. His abilities were very variable, ranging from extremely low in perceptual reasoning and working memory to borderline in verbal comprehension and processing speed. He is currently in the 5th grade, attends a special school without tests, and has an Individualized Education Program (IEP). When examined in-clinic, he could recall numbers 1–20 and had a concept of 'greater/less than'. His articulation was good, though he spoke in simple phrases. He was able to communicate his basic needs, though he had a low frustration tolerance. He was unable to perform simple calculations, and his verbal skills were at kindergarten levels. Additionally, he displayed a very childish and immature demeanor. His most recent IEP reported similar findings. Overall, this profile is supportive of the fact that his ability to communicate his symptoms was limited.

Seizures were first diagnosed at age two. Clinically, seizures consisted of bilateral arm elevation and extension with brief shaking for 3–5 s. At initial presentation, he had 15–20 seizures per day. Valproic acid significantly reduced his seizures and nearly abolished all clinical seizures within 2 months. Over the next few years, he continued to have frequent subclinical seizures on EEG, but no further clinical events occurred. The subclinical seizures consisted of buildup of 50- to 200-μV, rhythmic 4-Hz sharp waves over the mesial frontal region (maximal at Fz and Cz and also seen at Fp1, Fp2, F3, and F4), which would last for 35–50 s. These occurred solely in sleep, with no clinical correlation. Several medication trials were attempted to better control these events, and at the time of presentation, he was on a combination of valproic

acid, levetiracetam, and lacosamide. He has been on a variant of these three medications (though at varying doses) since the age of six. Further workup included a high resolution brain MRI, which showed no abnormalities. Psychiatrically, at the age of eight, he endorsed suicidal ideation. Given these psychiatric concerns in addition to excessive drowsiness in the setting of an improved EEG, his levetiracetam dose was reduced. Vitamin B6 was additionally started, and he began psychotherapy. This resulted in significant improvement in his mood and behavior.

He initially presented with his parents to his pediatrician and pediatric neurologist with complaints of odd new episodes. The episodes started after returning from a three-week vacation. During the episodes, the patient was noted to be restless, hyperactive, and upset. He would scratch his back and, at times, scream as well as run around the house. He complained that his back tickled and itched uncomfortably. These events were initially considered psychogenic based on the following: lack of complete stereotypy, no clinical seizures since the age of 2, history of behavioral problems, and the stresses associated with his parents enforcing limits on him. Additionally, given the presence of frontal epileptiform activity on prior EEGs, a sensory seizure did not appear to fit with the clinical symptoms.

Over time, his spells occurred with increasing frequency, happening multiple times per day including in association with arousal from sleep. He was seen in-clinic, and a decision was made to admit him for continuous video-EEG monitoring.

His cEEG showed frequent subclinical mesial frontal seizures, as he had exhibited previously. During the tickling events, there was initially so much movement it was not possible to tell whether these were behavioral or ictal in nature. Behavioral management to help him relax was provided. Additionally, he was started on clonidine at night to aid with sleep and to medically treat the outbursts. This resulted in improvement in the level of distress of the patient and the family, as the behaviors interfered less with the patient's daily functioning, though the frequency of the tickling attacks remained unchanged.

After these modifications reduced the emotional upset and hyperactivity associated with the tickling attacks, the stereotyped nature of these events became more apparent and the ictal EEG features more discernable. The patient reliably complained of itching of the right middorsal region, which suggested a more posterior focus than was apparent on EEG. To better define the potential epileptogenic and symptomatogenic zones, double-density electrodes were placed over the bilateral frontal and parietal head regions. He had numerous events per day and, at times, numerous events per hour, resulting in an EEG that showed 130- to 180-μV spikes and sharp waves over the left frontocentral region (FC3 > F3, Fz, FCz) which built up into rhythmic, sharply contoured slowing (1–4 Hz, 200–300 μV) over the left frontocentral region (Fp1, F3, C3). At times, this activity would propagate to the right frontocentral region (Fp2, F4, C4) as well. Clinically, the patient cried out "it tickles" consistently. Occasionally, right shoulder jerking or rubbing/scratching of the right side of his back were associated with his cries. These runs of epileptiform discharges were only seen during the tickling attacks, thus confirming them as true ictal phenomena.

4. Discussion

We theorize that our patient has a deep epileptic generator that is not visible electrographically at the ictal onset with standard scalp electrodes but is only visible on EEG after it propagates to the frontal lobe. His symptomatogenic zone involves the left somatosensory cortex and manifests as recurrent tickling attacks. Given his age, developmental delays, and very childlike personality, further characterization of the sensation he feels was not possible. Additionally, it is possible that the epileptogenic generator is located within a gyrus at an angle that can only be seen on EEG in the frontal region because of its orientation. Magnetoencephalography (MEG) could help to clarify this. Subtracted ictal spect coregistered to MRI brain (SISCOM) could also help improve localization of his seizure onset. Without a focal structural lesion identified by high resolution brain MRI or abnormality found with cortical thickness analysis, as well as likely involvement of eloquent cortex, the patient is not felt to be a surgical candidate; thus, these studies have not been completed.

There have been many advances in our ability to treat epilepsy as well as PNES in the past decade, and video-EEG continues to be the gold standard in distinguishing these two entities. Recent studies have demonstrated the efficacy of cognitive–behavioral therapy (CBT) as well as selective serotonin reuptake inhibitors (SSRIs) and, in particular, the combination of the two in combating PNES [5]. Though, clearly the first step is excluding true epilepsy.

Parietal lobe seizures are by far the least studied of focal onset seizures and may be particularly challenging to diagnose in children because of the subjective nature of associated sensory phenomena. Patients with parietal lobe seizures manifest symptoms in a wide variety of ways and not solely with simple sensory phenomena [6]. Reports in the literature as to the semiology of parietal seizures vary greatly, including the following: staring, tonic posturing, motor weakness, sensory changes, eye deviation [7], and rarely, focal pain [8].

In our patient, his seizure manifested as a right posterior trunk unpleasant sense of ticklishness. These seizures were clearly uncomfortable, but his developmental disability precluded parsing whether the discomfort was truly pain. Epileptic pain is uncommon, and diagnosis of epileptic seizures in patients with epileptic pain is frequently delayed or inaccurate initially [9]. Additionally, the behavioral overlay our patient displayed made diagnosis of these events as epileptic in nature significantly more difficult. In children, particularly those with developmental delays, behavioral reactions are common as the child is unable to communicate his/her symptoms as eloquently as most adults.

This case of new-onset 'tickling' seizures emphasizes the challenges of diagnosing parietal lobe seizures in children. It affirms the importance of using psychotropic medications, behavioral modifications, and cEEG monitoring, with double-density electrodes in specific instances to make the correct diagnosis.

Author contributions

Dr. Jessica Falco-Walter drafted/edited the manuscript and performed analysis and interpretation of the data. Dr. Michael Stein and Dr. Maggie McNulty edited the manuscript and performed analysis and interpretation of the data. Dr. Lubov Romantseva and Dr. Peter Heydemann edited the manuscript and provided supervision.

Appendix A. Supplementary data

Supplementary data to this article can be found online at http://dx.doi.org/10.1016/j.ebcr.2016.07.002.

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