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CASE REPORT

A Case of Volvular Epileptic Seizure Associated with Intracranial Cortical Lesion

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Abstract

Volvular epileptic seizure is characterized by walking repetitively in small circles and it is a rare kind of seizure. It is a result of focal epileptic activity. It could coexist with adversive seizures but it is considered as a separate seizure semiology. Volvular seizures may occur due to primary epileptic syndrome or secondary to an intracranial lesion. Volvular seizures are mostly seen in frontal lobe lesions, whereas it can be associated with temporal, parietal, or occipital lesions. We report a case who has volvular epilepsy started at age of 55 due to an intracranial lesion which we considered and treated as an autoimmune lesion and controlled his resistant epilepsy. New onset epileptic seizures can be caused by autoimmune etiology.

Keywords: Autoimmune epilepsy; volvular epilepsy; volvular seizure.

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Introduction

Volvular epilepsy is characterized by walking repetitively in small circles and it is a rare kind of seizure. It is a result of focal epileptic activity. It could coexist with adversive seizures but it is considered as a separate seizure semiology. We report a case who has volvular epilepsy due to intracranial lesion.

Case Report

A 55-year-old male patient presented with focal clonic seizure in his left arm and leg lasting approximately 1–2 min repeating 20–30 times a day. Seizure onset was a month ago. He also had focal impaired awareness seizures characterized by losing awareness and staring blankly which started 2 days ago and negative myoclonus in his left arm and leg triggered by voluntary movements. His full blood count, liver, and kidney function tests, electrolytes were in normal range. His cranial magnetic resonance imaging (MRI) revealed dural contrast enhancing lesion at right frontoparietal region (Fig. 1b). Interictal electroencephalography (EEG) showed sharp waves in the right frontocentroparietal region (Fig. 1b). During video EEG monitorization, the patient had focal motor epileptic seizure characterized by clonic movements in his left arm and leg. EEG showed rhythmic activity at the right frontocentroparietal region during this focal motor seizure. Seizure frequency was reduced with 3000 mg/day levetiracetam but full seizure control was not established. Lumbar puncture revealed no cells; cerebrospinal fluid (CSF) protein, glucose, and pressure were normal. Oligoclonal bands were negative, and IgG index was normal (0.43). CSF culture revealed no bacterial growth and CSF tuberculosis polymerase chain reaction test was negative. Autoimmune encephalitis panel (including anti NMDA Ab, AMPA-R1 Ab, AMPA-R2 Ab, CASPR2 Ab, LGI1 Ab, and GABA-R Ab) and paraneoplastic panel (including anti-Hu, anti-Yo, anti-Ri, anti-amphiphysin, anti-Tr, anti-PCA-2, anti-Ma, anti-CV2 1, and anti-ANNA 3 Ab) were negative. Serum angiotensin converting enzyme (ACE) was normal, only CSF ACE was high (7.53, reference <2.5). Thorax computerized tomography revealed emphysema in

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İntrakraniyal Kortikal Lezyona Bağlı Volvüler Epileptik Nöbet Olgusu

Öz

Volvüler epileptik nöbet hastanın bir çember çizerek yürümesiyle karakterizedir ve nadir görülen bir nöbet çeşididir. Fokal epileptik aktivite sebebiyle ortaya çıkar. Adversif nöbetlerle beraber olabilir ancak ayrı bir nöbet semiyolojisi olarak değerlendirilir. Primer epileptik sendroma bağlı olarak ya da intrakraniyal lezyona sekonder olarak ortaya çıkabilir. Volvüler nöbet, çoğunlukla frontal lob lezyonlarına bağlı ortaya çıkmakla beraber; temporal, parietal ya da oksipital lob lezyonlarına bağlı da gelişebilir. Bu yazıda, 55 yaşında intrakraniyal lezyona bağlı volvüler epileptik nöbetleri başlayan, altta yatan sebebi otoimmünite olarak değerlendirerek dirençli nöbetlerini kontrol altına aldığımız bir olgunun sunulması amaçlanmıştır. Yeni başlangıçlı nöbetlerin altında otoimmün etiyoloji yatabilmektedir.

Anahtar sözcükler: Otoimmün epilepsi; volvüler epilepsi, volvüler nöbet.

the upper lobes and pleural thickenings. There was no hilar lymphadenopathy, no hypercalciuria in 24 h urine sample. Mediastinal lymph node biopsy was normal. Positron emission tomography scan revealed no sign of malignity. Our investigations covering infections, malignities, vasculitic diseases, and sarcoidosis were negative. Although the autoimmune encephalitis panel was negative, we accepted that the lesion may have an autoimmune origin after ruling out other potential causes and started 1 gr/day intravenous methylprednisolone. After 7 days of treatment, he became seizure free and discharged with levetiracetam 3000 mg/day. After a month, he presented with volvular seizures repeating every hour for last 4 days. He gets up and walks in circles turning towards left and does not answer questions while walking, he completes 2–3 circles and falls after circling, which was evaluated as volvular epileptic seizure. Unfortunately, the patient did not have any volvular seizure during EEG recording; therefore, we could not monitor ictal EEG pattern of his volvular epileptic seizures. Seizures got under control after 1000 mg/day valproic acid was added to his treatment. Biopsy was planned if there is enlargement at the lesion size at consequent MRI scans. Control cranial MRI after 1 month showed slight regression (Fig. 2a), control MRI after 6 months showed significant regression at dural contrast enhancement (Fig. 2b).

Informed consent was obtained from the patient to use his medical information in a case report to be shared by healthcare professionals.

Discussion

Epileptic seizures are often associated with paraneoplastic and autoimmune syndromes.[1] Dubey et al.[2] reported a series of 127 patients with epilepsy of unknown origin and 20.5% of patients presented antibodies that strongly implied an autoimmune origin of this disease. International League against Epilepsy also included epilepsy of autoimmune origin to its classification recently.[3]

Autoimmune epilepsy has to be taken into consideration in cases with new onset seizures. It was also shown autoimmune and paraneoplastic etiology is responsible for

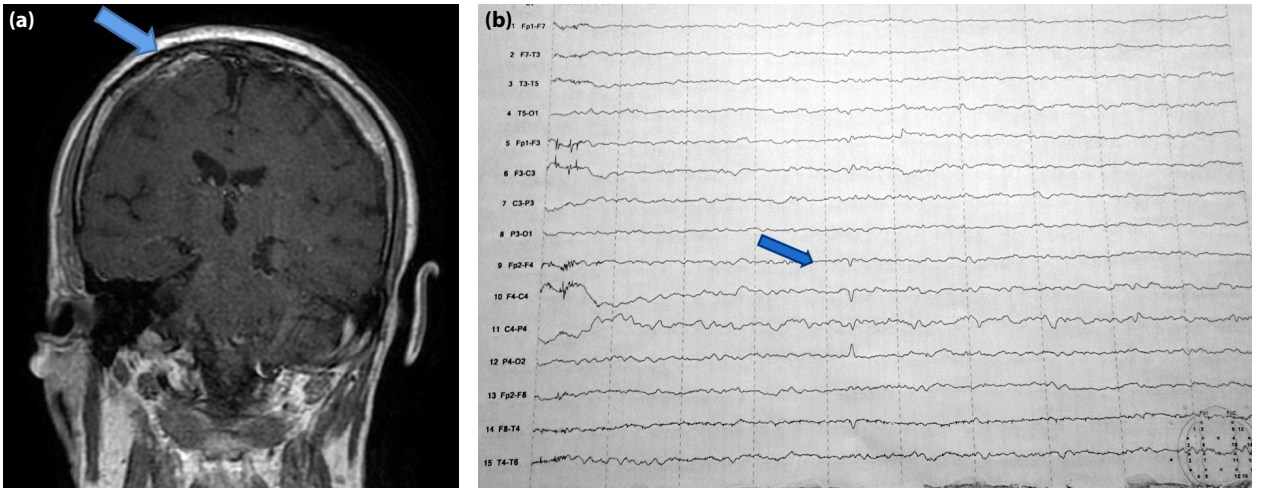

Fig. 1. **(a)** Dural contrast enhancing lesion at the right frontoparietal region (magnetic resonance imaging). **(b)** Sharp waves at the right frontocentroparietal region (electroencephalography).

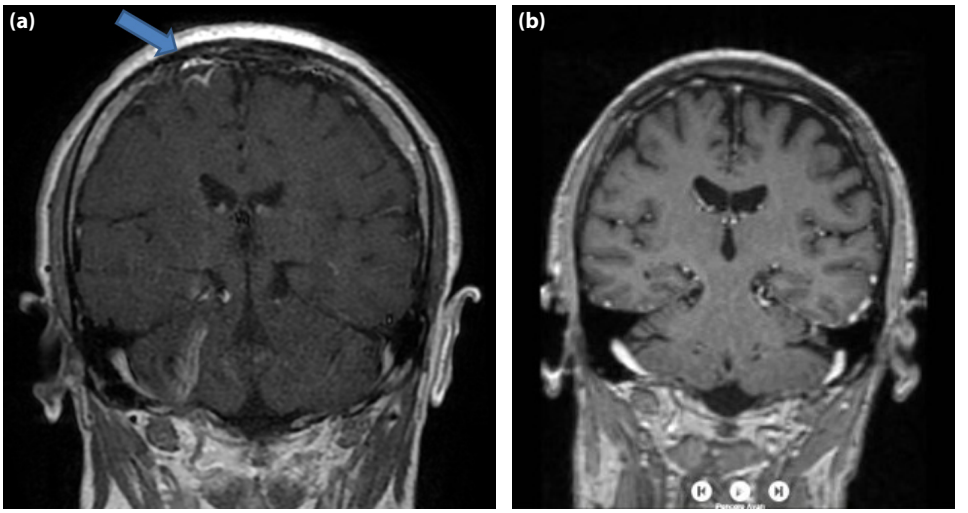

Fig. 2. **(a)** Slight regression at contrast-enhanced lesion after a month (magnetic resonance imaging). **(b)** Significant regression at contrast-enhanced lesion after 6 months (magnetic resonance imaging).

13–37% of newly occurring, unprovoked, and refractory status epilepticus cases.[4] Autoantibodies cannot always be demonstrated in these cases. Sensitivity of antibody testing was found to be low (60–80%) in clinically defined autoimmune encephalitis.[5] We have also diagnosed and treated our case as autoimmune encephalitis after ruling out other possible diagnosis, despite antibodies were negative.

Seizures with autoimmune etiology are known to be resistant to antiepileptic drugs but responsive to immunotherapy, as seen in our case. A study assessing electroclinical features of 19 patients who have seizures due to autoimmune encephalitis, reported that most frequent seizure form was subclinic seizures among these patients. Focal seizures with preserved awareness were seen in 4% of the patients. Volvular seizure was not documented in any of the cases.[6]

Versive seizures are well known which are characterized by head and body turning in a horizontal axis. There are many articles related to head and body turning during epileptic seizure in the literature whereas volvular epileptic seizure characterized by walking in small circles is a rare kind of seizure.

Alqadi et al.[7] stated that 10 out of 116 patients who have undergone epilepsy surgery and have hypermotor seizures between 1996 and 2013, had nonversive head and body turning (ranging from 90° to 270°). Turning was ipsilateral to the side of resection in all patients and seen both in frontal and extrafrontal resections according to their research. On the other hand, Leung et al.[8] reported that ictal body turning was localizing to the mesial frontal region, and this would be helpful for distinguishing mesial frontal epilepsy from other frontal epilepsy entities such as lateral frontal lobe epilepsy and orbitofrontal lobe epilepsy. Ictal body turning occurred in 16 patients among 28 patients who had mesial frontal lobe epilepsy whereas it only occurred in 13 patients among 200 patients with nonmesial frontal epilepsy according to their research. They also proposed that turning could be ipsilateral or contralateral in equal proportions.Another study proposed that according to their study, all of the temporal lobe epileptic seizures had both contralateral and ipsilateral head turning, whereas all frontal lobe epileptic seizures had contralateral head turning; only six of 22 seizures were associated with ipsilateral head turning. Ipsilateral head turning always preceded contralateral head turning in both temporal and frontal lobe epilepsies.[9] Mercan et al.[10] stated that nonversive body turning ipsilateral to the epileptic zone and Versive body turning contralateral to the epileptic zone is valuable at lateralization in temporal and extratemporal lobe epileptic seizures, especially when present with head turning. They also stated that in temporal lobe epileptic seizures, both Versive body turning and nonversive body turning are probably caused by two different pathophysiological mechanisms similarly to head turning. Versive body turning seems to be related with cortical area spreading, whereas nonversive body turning is more related with basal ganglia involvement.

Versive seizures are not uncommon and sometimes truncal rotation happens as well as head turning, resulting in turning. Walking in circles during a seizure is different because

walking is a complex integrated motor activity. Donaldson reported four cases with volvular seizures whose epileptic foci were at anterior frontal, temporal, parietal, and occipital regions. Therefore, he stated that volvular seizures seem to originate from deeper structures such as striatum (caudate, putamen, and globus pallidus complex), rather than cortex. There are several reported cases where direction of rotation is toward the lesion or away from the lesion, which means, direction of rotation does not have lateralizing significance. Walking in circles is a focal epileptic activity that can remain localized or can be followed by a secondarily generalized epileptic seizure.[11] Our patient with right frontocentroparietal lesion was rotating toward left while walking, contralateral to the epileptic zone.

Conclusion– Volvular epileptic seizure which is a rare kind of seizure may occur due to primary epileptic syndrome or secondary to an intracranial lesion. Volvular seizures are mostly seen in frontal lobe lesions, whereas it can be associated with temporal, parietal or occipital lesions. Involvement of basal ganglia could be the underlying mechanism of circling.

Informed Consent– Written informed consent was obtained from patients who participated in this study.

Peer-review– Externally peer-reviewed.

Authorship Contributions– Concept: F.E., G.G.; Design: F.E., G.G.; Supervision: F.E., E.Ç.; Data collection &/or processing: E.Ç., H.C.S.; Analysis and/or interpretation: A.S., F.E.; Literature search: H.C.S.; Writing: H.C.S.; Critical review: F.E., A.S.

Conflict of interest– The authors declare that they have no conflict of interest.

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