

Delusional Pregnancy in a Patient with Epilepsy: A Case Report

Epilepsi Tanılı Hastada Gebelik Sanrısı: Bir Olgu Sunumu

Ömer Naim SAYER

0000-0001-8104-6219

Çiçek HOCAOĞLU

0000-0001-6613-4317

Department of Psychiatry, Recep
Tayyip Erdoğan University Faculty of
Medicine, Rize, Türkiye

ABSTRACT

Interictal psychosis is a psychotic symptom that is not temporally related to epileptic seizures. Pregnancy delusion is defined as a person's fixed belief that she is pregnant despite objective evidence that she is not pregnant. In this case report, pregnancy delusion was described in a patient with epilepsy. A 31-year-old woman with epilepsy was admitted to a psychiatric ward. The patient, whose pregnancy test results were never positive, believed that she was pregnant. It was learned that her identical twin had experienced reproductive-sexuality-themed psychotic symptoms 10 years ago. The patient was hospitalized for three weeks and discharged in remission with paliperidone 6 mg/day and biperiden 2 mg/day. Caution should be exercised when using antipsychotics because of their epileptogenic effects. Pregnancy delusion in epilepsy is rare. In addition to this rare condition, it is noteworthy that reproductive-sexual delusions were reported in the patient's twin brother who was diagnosed with epilepsy.

Keywords: Pseudocyesis; epilepsy; psychosis.

ÖZ

İnteriktal psikoz, epileptik nöbetlerle zamansal olarak ilişkili olmayan psikotik bir semptomdur. Gebelik sanrısı, bir kişinin hamile olmadığına dair nesnel kanıtlara rağmen hamile olduğuna dair sabit inancı olarak tanımlanır. Bu vaka raporunda, epilepsili bir hastada gebelik sanrısı tanımlanmıştır. Otuz bir yaşında epilepsi hastası bir kadın psikiyatri servisine yatırılmıştır. Gebelik test sonuçları hiçbir zaman pozitif çıkmayan hasta gebe olduğuna inanıyordu. Hastanın tek yumurta ikizinin 10 yıl önce üreme-cinsellik temalı psikotik belirtiler yaşadığı öğrenildi. Üç hafta hastanede yatan hasta paliperidon 6 mg/gün ve biperiden 2 mg/gün tedavisiyle remisyonla taburcu edilmiştir. Epileptojenik etkileri nedeniyle antipsikotik kullanırken dikkatli olunmalıdır. Epilepside gebelik sanrısı nadirdir. Bu nadir duruma ek olarak, hastanın epilepsi tanısı almış ikiz kardeşinde de üreme-cinsel sanrıların bildirilmiş olması dikkat çekicidir.

Anahtar kelimeler: Yalancı gebelik; epilepsi; psikoz.

INTRODUCTION

Epilepsy stands as one of the most common chronic neurological diseases worldwide, with a reported prevalence of 0.4-1% (1). Psychiatric disorders frequently accompany epilepsy, with studies indicating that 39-54.1% of diagnosed patients exhibit such comorbidities (2). A systematic review revealed that around 6% of individuals diagnosed with epilepsy -eight times more than the general population- experience psychotic disorders (3).

Corresponding Author

Sorumlu Yazar

Ömer Naim SAYER
omernaimsayer@gmail.com

Received / Geliş Tarihi : 22.01.2024

Accepted / Kabul Tarihi : 22.05.2024

Available Online /

Çevrimiçi Yayın Tarihi : 10.08.2024

Presented as a poster at the 8th Psychiatry Summit and 15th Anxiety Congress (May 18-21, 2023; KKTC).

Factors such as the onset of epilepsy before the age of ten and temporal lobe origin increase the risk of developing psychosis. Additionally, there is a suggestion that controlling seizures and the use of antiepileptic drugs may heighten the risk of psychosis. In this context, the concept of 'forced normalization,' a significant phenomenon in the relationship between psychiatric disorders and epilepsy, deserves attention (4). Two main hypotheses attempt to explain the relationship between epilepsy and psychosis. The first suggests that recurrent epileptic seizures predispose individuals to psychosis due to their neurotoxic effect, while the second posits that epilepsy and psychosis result from common neurodevelopmental disorders or nonspecific diffuse brain damage (5). Supporting the second hypothesis, neuropathological, neuroimaging, and genetic findings indicate similarities in structural brain abnormalities and genetic irregularities between patients with schizophrenia and epilepsy (3).

Psychotic symptoms in epilepsy patients are categorized as ictal, post-ictal, and interictal. Interictal psychosis, not associated with seizures, generally exhibits a clinical appearance similar to schizophrenia (2). Unlike schizophrenia, interictal psychosis presents more prominent positive symptoms, with less impairment in cognitive functions and overall functionality (6).

Pregnancy delusion, classified as a somatic delusional disorder within the schizophrenia spectrum and other psychotic disorders in the Diagnostic and Statistical Manual of Mental Disorders-5 (DSM-5), involves a fixed belief in pregnancy despite clear objective evidence to the contrary. Unlike pseudocyesis, pregnancy delusion lacks physical symptoms of pregnancy (7,8). The etiology of pregnancy delusion and pseudocyesis involves various biological, social, and psychological factors. Both conditions are rare, with limited literature available on their occurrence in patients diagnosed with epilepsy (8). This case report aimed to contribute to the existing literature by discussing the interictal psychosis observed in a patient diagnosed with epilepsy, emphasizing the central theme of pregnancy delusion.

CASE REPORT

A 31-year-old female patient, a high school graduate, and housewife, second among five siblings, and an identical twin, has been evaluated for compulsory hospitalization at the psychiatry outpatient clinic, accompanied by her father. Her husband, from whom she has been separated for two months, applied to the court, citing the patient's psychiatric symptoms. The court issued a 'decision to hospitalize the patient in the psychiatric ward.' During the interview, accusatory statements by both the patient and her father against her husband were prominent. The patient claimed her husband sought hospitalization to facilitate divorce, denying any psychiatric illness. She also disclosed a terminated pregnancy due to violence from her husband five months ago. Medical records revealed a previous diagnosis of bipolar disorder, but the patient hadn't adhered to the recommended treatment.

Further examination of medical records revealed consistently negative human chorionic gonadotropin (hCG) values despite frequent pregnancy test visits. The family physician noted the patient's insistence on blood tests, doubting negative results. The patient claimed to have seen

the gestational sac in an ultrasound but believed doctors were concealing it.

In her medical history, the patient had been treated for focal epileptic seizures since childhood, with the last seizure occurring two years ago. Currently on lacosamide 150 mg 2*1, the patient's last psychiatric treatment, olanzapine 2.5 mg, was discontinued due to sleepiness. Compulsory hospitalization led to her admission for organized treatment. Routine blood tests, brain magnetic resonance imaging, and electroencephalography were normal.

The patient's parents, both teachers, had no known psychiatric diagnosis but shared their daughter's pregnancy delusions, asserting her lack of psychiatric illness. The patient's identical twin, also diagnosed with epilepsy, had been hospitalized in a psychiatric ward a decade ago. Born prematurely, the twin experienced the first epileptic seizure at 55 days and exhibited psychiatric symptoms ten years ago. Despite admission to psychiatry, she left the clinic prematurely at her family's request without completing treatment.

Mental State Examination

The individual appeared to be her stated age, appropriately dressed for her sociocultural level, displayed good self-care, was slightly overweight, maintained eye contact, responded to questions purposefully, and exhibited a normal speech rate and amount. She presented a defensive attitude, affective irritability, and a dysphoric mood. Consciousness was clear, and she demonstrated orientation, and cooperation, with no psychopathological findings in perception. Her intelligence level was clinically normal, abstract thinking ability was intact, but there was an impairment in her ability to evaluate reality. The content of her thoughts included delusions of seeing evil in her husband and a delusion of pregnancy. Psychomotor agitation was observed.

Clinical Course

Hospitalized with a DSM-5 diagnosis of delusional disorder, the patient actively participated in clinic and garden activities. Psychometric evaluations indicated scores of 17 points on the young mania rating scale (YMRS), 22 points on the brief psychiatric rating scale (BPRS), 21 points on the scale for the assessment of positive symptoms (SAPS), and 2 points on the scale for the assessment of negative symptoms (SANS).

In interviews, accusatory and suspicious thoughts towards her husband were prominent. She reported frequent visits to the gynecology outpatient clinic, claiming to have seen a gestational sac on ultrasonography, contrary to physicians' findings. Risperidone 1 mg/day treatment was initiated and gradually increased. Considering the patient's epilepsy diagnosis, a consultation with the neurology unit recommended continuing the current epilepsy treatment (lacosamide 150 mg 2*1).

On the third day, the patient reported body rashes and refused medication, though no dermatologic lesion was found on examination. Non-compliant attitudes towards risperidone were noted in the patient and her parents, leading to a decision to reorganize treatment. Considering prolonged-release formulations for better compliance, paliperidone 3 mg/day was initiated. The patient remained compliant, exhibited no issues with peers, and expressed a desire to study medicine. Accusatory speeches against her

husband continued during occasional doctor visits, where she alleged violence resulting in a miscarriage, unsupported by physical examination findings. The patient adhered to paliperidone treatment, with the dose increased to 6 mg/day. Serum prolactin levels were monitored, and repeated psychometric tests showed symptom improvement (YMRS: 4, BPRS: 14, SAPS: 6, SANS: 0 points). Outpatient treatment was decided, and the patient was discharged on the 22nd day with follow-up plans. Informed consent was obtained from the patient and her family, who continue regular outpatient follow-up.

DISCUSSION

Psychotic symptoms associated with epilepsy are not clearly categorized in DSM-5, leading to difficulties in diagnosis (3). The bidirectional relationship between epilepsy and psychiatric disorders may pose challenges in clinical practice (9). These patients may present with various clinical manifestations, leading to potential misdiagnosis. In fact, our patient's past diagnosis of bipolar disorder supports this observation. The less impaired functionality in interictal psychosis, the prominence of positive symptoms, and the onset of epilepsy preceding psychosis may aid in distinguishing it from primary psychotic disorders (6).

Studies indicate a poor prognosis for interictal psychoses, with a small proportion recovering spontaneously, while approximately two-thirds persist beyond six months (10,11). While no specific guidelines exist for managing interictal psychoses, applying treatment schemes for initial psychotic episodes is recommended. Besides using antiepileptic drugs to control seizures, using antipsychotic drugs for a period has been emphasized for managing psychotic symptoms (10).

In our case, the addition of antipsychotic drugs to antiepileptic treatment resulted in regression of psychotic symptoms. Choosing an antipsychotic drug for interictal psychosis lacks a clear answer, necessitating caution due to the epileptogenic effects of these drugs. Particularly, chlorpromazine and clozapine are well-known to lower seizure thresholds (12).

Some authors argue against distinguishing between the delusion of pregnancy and pseudocyesis, suggesting their continuous nature (13). Consequently, disorders in the pseudocyesis differential diagnosis should also be considered in delusions of pregnancy. Although many neurological, endocrine, and metabolic diseases are implicated in this delusion, the association with epilepsy is not commonly emphasized (13). Interestingly, a case similar to ours, involving delusions of pregnancy in a young female diagnosed with epilepsy, has been reported, necessitating further studies to unveil the relationship between epilepsy and delusions of pregnancy (14).

In the literature, it is noted that patients with pregnancy delusions provide responses aligned with their sociocultural level, contrary to medical evidence (10). In our case, the patient claimed to recognize the gestational sac on USG, attributing it to her high school child development studies, challenging the doctors' findings.

Reproductive and sexuality-themed delusions accompanying pregnancy delusion have been suggested (13). Similarly, it is intriguing that the identical twin of our patient developed such delusions. Both siblings, treated for epilepsy since

infancy, suggest common neurobiological processes in the pathogenesis.

Our case revealed that the patient's parents also shared the delusion of pregnancy, resembling a rare instance of shared psychosis syndrome known as "Folie-à-deux," where delusions transfer to another person in a close relationship. A reported case in the literature involves a married couple sharing the delusion of pregnancy (15). The involvement of the patient's relatives in psychosis complicates treatment as family support becomes challenging to obtain.

In summary, key points from our case report include the absence of appropriate classification and diagnostic criteria for epilepsy-related psychotic symptoms, the potential for misdiagnosis due to varied clinical presentations in interictal psychosis, and the occurrence of delusions of pregnancy as part of interictal psychosis. The development of reproductive-sexuality-themed delusions in the patient's identical twin, both treated for epilepsy since infancy, hints at common neurobiological processes in the pathogenesis. Pregnancy delusions might extend beyond the individual, impacting social life.

In conclusion, the need for guidelines in diagnosing and treating interictal psychosis is evident. Further neurobiological studies exploring the relationship between epilepsy and pregnancy delusion are crucial. We believe our study can raise awareness among clinicians regarding this subject.

Informed Consent: Written informed consent was obtained from the patient for publication.

Conflict of Interest: None declared by the authors.

Financial Disclosure: None declared by the authors.

Acknowledgments: None declared by the authors.

Author Contributions: Idea/Concept: ÖNS, ÇH; Design: ÖNS, ÇH; Data Collection/Processing: ÖNS; Analysis/Interpretation: ÖNS, ÇH; Literature Review: ÖNS; Drafting/Writing: ÖNS, ÇH; Critical Review: ÇH.

REFERENCES

- Swinkels WA, Kuyk J, van Dyck R, Spinhoven P. Psychiatric comorbidity in epilepsy. *Epilepsy Behav.* 2005;7(1):37-50.
- Irwin LG, Fortune DG. Risk factors for psychosis secondary to temporal lobe epilepsy: a systematic review. *J Neuropsychiatry Clin Neurosci.* 2014;26(1):5-23.
- Clancy MJ, Clarke MC, Connor DJ, Cannon M, Cotter DR. The prevalence of psychosis in epilepsy; a systematic review and meta-analysis. *BMC Psychiatry.* 2014;14:75.
- Bahali MK, Zengin M, Tamam L, Ozpoyraz N, Bozdemir H. Forced normalization: an important phenomenon in the relationship between psychiatric disorders and epilepsy. *Psychiatry Clin Psychopharmacol.* 2008;18(4):322-6.
- Sachdev P. Schizophrenia-like psychosis and epilepsy: the status of the association. *Am J Psychiatry.* 1998;155(3):325-36.

6. Tascon-Cervera JJ, Crisostomo-Siverio AI, Cardenes-Moreno C, Dorta-Gonzalez JF, Morera-Fumero AL. Psychosis in epilepsy vs late-onset schizophrenia: a case report. *Cureus*. 2022;14(12):e32692.
7. Ray M, Kumar S, Sinha A, Pisharody RR. Delusion of pregnancy: conception in brain. *Ind Psychiatry J*. 2022;31(2):341-5.
8. Bera SC, Sarkar S. Delusion of pregnancy: a systematic review of 84 cases in the literature. *Indian J Psychol Med*. 2015;37(2):131-7.
9. Gurgu RS, Ciobanu AM, Danasel RI, Panea CA. Psychiatric comorbidities in adult patients with epilepsy (A systematic review). *Exp Ther Med*. 2021;22(2):909.
10. Adachi N, Kanemoto K, de Toffol B, Akanuma N, Oshima T, Mohan A, et al. Basic treatment principles for psychotic disorders in patients with epilepsy. *Epilepsia*. 2013;54(Suppl 1):19-33.
11. Adachi N, Akanuma N, Ito M, Okazaki M, Kato M, Onuma T. Interictal psychotic episodes in epilepsy: duration and associated clinical factors. *Epilepsia*. 2012;53(6):1088-94.
12. Henning O, Nakken KO. Epilepsirelaterte psykoser [Epilepsy-related psychoses]. *Tidsskr Nor Laegeforen*. 2013;133(11):1205-9. Norwegian.
13. Seeman MV. Pseudocyesis, delusional pregnancy, and psychosis: the birth of a delusion. *World J Clin Cases*. 2014;2(8):338-44.
14. Paholpak P, Gan JJ, Mendez MF. Neuropsychiatric aspects of epilepsy. In: Sadock BJ, Sadock VA, Ruiz P, editors. *Kaplan and Sadock's comprehensive textbook of psychiatry*. 10th ed. Philadelphia: Wolters Kluwer; 2017. p.517.
15. Nagdive AB, Bhainsora RS, Fernandes R, Behere PB, Sethi S. Pseudocyesis leading to folie-à-deux. *J Neurosci Rural Pract*. 2021;12(2):419-23.