Cumhuriyet Medical Journal, 44(4): 484-488, 2022
DOI: http://dx.doi.org/10.7197/cmj.1194080

Cumhuriyet Medical Journal

| cmj.cumhuriyet.edu.tr | Founded: 2004 Available online, ISSN:1305-0028 Publisher: Sivas Cumhuriyet Üniversitesi

Sleep-Isolated Trichotillomania As An Ictal Symptom Of Complex Partial Seizure In A 12-Year-Old Girl: A Case Report

Beyza Bozok[1], Öykü Özdemir[1], Ayla Uzun Çiçek[1],*, Halil Ural Aksoy[2], İlknur Ucuz[3], Elif Abanoz[4]

[1]Department of Child and Adolescent Psychiatry, Faculty of Medicine, Sivas Cumhuriyet University, Sivas, Turkey, [2]Child Neurology Clinic, Manisa City Hospital, Manisa, Turkey, [3]Department of Child and Adolescent Psychiatry, Faculty of Medicine, İnönü University, Malatya, Turkey, [4]Child and Adolescent Mental Health and Diseases Clinic, Elazığ Mental Health and Diseases Hospital, Elazığ, Turkey

**Corresponding author*

Case Report

History

Received: 26/10/2022
Accepted: 25/12/2022

ABSTRACT

Sleep-isolated trichotillomania (TTM) is the unconscious and automatic pulling of one's hair only while sleeping. In previous case reports, sleep-isolated TTM has been categorized as a sleep-related disorder or a sleep-related dissociative disorder in case reports, and a treatment strategy in this direction has been implemented. However, to our knowledge, no reports have been published describing hair pulling during sleep as a symptom of epilepsy and its pharmacological treatment. In this case report, we present the hair-pulling behavior during sleep as an ictal manifestation of complex partial seizure in a 12-year-old girl and its successful treatment with levetiracetam. We suggest that epilepsy should be suspected in addition to parasomnias and sleep-related dissociative disorder in the differential diagnosis, especially in cases presenting with isolated TTM behavior during sleep.

Keywords: Trichotillomania, sleep, sleep-isolated trichotillomania, epilepsy, levetiracetam

12 Yaşında Bir Kız Çocuğunda Kompleks Parsiyel Nöbetin İktal Bir Semptomu Olarak Uykuda İzole Trikotillomani: Bir Olgu Sunumu

Süreç

Geliş: 26/10/2022
Kabul: 25/12/2022

ÖZ

Uykuda izole trikotillomani (TTM), kişinin sadece uyurken bilinçsiz ve otomatik olarak saçını koparmasıdır. Daha önceki vaka bildirimlerinde, uykuda izole TTM, uyku ile ilişkili bir bozukluk veya uyku ile ilişkili dissosiyatif bozukluk olarak kategorize edilmiş ve bu yönde bir tedavi stratejisi uygulanmıştır. Bununla birlikte, bildiğimiz kadarıyla, epilepsinin bir semptomu olarak uykuda saç yolma ve farmakolojik tedavisini açıklayan hiçbir rapor yayınlanmamıştır. Bu olgu sunumunda 12 yaşındaki bir kız çocuğunda kompleks parsiyel nöbetin iktal bir belirtisi olarak uykuda saç yolma davranışı ve bunun levetiresetam ile başarılı tedavisini sunuyoruz. Özellikle uykuda izole TTM davranışı ile başvuran olgularda ayırıcı tanıda parasomniler ve uyku ile ilişkili dissosiyatif bozukluk yanı sıra epilepsi varlığından da şüphelenilmesi gerektiğini öneriyoruz.

Anahtar sözcükler: Trikotilomani, uyku, uykuda izole trikotilomani, epilepsi, levetiresetam

[a] drbeyza.44@gmail.com https://orcid.org/0000-0003-4205-5360
[b] oykuozdemir06@gmail.com https://orcid.org/0000-0001-5253-1039
[c] dr.f.ayla@hotmail.com https://orcid.org/0000-0003-2274-3457
[d] uralaksoy@hotmail.com https://orcid.org/0000-0003-3131-1808
[e] ilknur_27@yahoo.com https://orcid.org/0000-0003-1986-4688
[f] elifabanoz_17@hotmail.com https://orcid.org/0000-0002-9214-4735

How to Cite: Bozok B, Özdemir Ö, Uzun Çiçek A, Aksoy HU, Ucuz İ, Abanoz E (2022) Sleep-Isolated Trichotillomania As An Ictal Symptom Of Complex Partial Seizure In A 12-Year-Old Girl: A Case Report, Cumhuriyet Medical Journal, December 2022, 44 (4): 484-488

Introduction

Trichotillomania (TTM) is a psychiatric disorder that causes noticeable hair loss as a result of repetitive hair-pulling behavior [1]. TTM is thought to occur with the interaction of multiple factors including genetic and familial, neurobiological, environmental, and psychological variables [2-4]. The differential diagnosis of TTM includes dermatological conditions, hormonal causes, drug side effects, nutritional conditions, and psychiatric conditions, especially body dysmorphic disorder and obsessive-compulsive and related disorders [2-4]. TTM treatment is a combination of behavioral and psychopharmacological interventions [4, 5].

In TTM, there are two types of hair-pulling behavior as focused and automatic. In focused pulling behavior, the hair is consciously plucked. There is an irresistible urge to pull out the hair and relief is provided when the hair is pulled out. In automatic behavior, the patient is not aware of the picking behavior, the behavior is unconscious or automatic. These patients may pull their hair out while watching TV, reading a book, or when they are bored [2-4, 6]. An extreme example of automatic TTM is that some patients only pull their hair while asleep, and this is called sleep-isolated TTM [6, 7]. There are very few case reports and only one study regarding sleep TTM behavior [6-11]. Again, there is only one case report showing that TTM is the behavior that occurs as a symptom of epilepsy, but in this case report, TTM behavior was observed while awake [12]. In fact, another case of a 14-year-old severely intellectually disabled autistic girl presenting with aggressive outbursts, trichotillomania, and atypical minor epileptic seizures has been reported, but in this case report, trichotillomania has only been mentioned as an incidental finding and no detailed information on its occurrence or treatment was given [13]. To our knowledge, no reports have been published describing hair pulling during sleep as a symptom of epilepsy, and its pharmacological treatment. In this case report, trichotillomania during sleep, which was detected as the ictal symptom of complex partial seizure in a 12-year-old girl, and its successful treatment with levetirecetam is described. Written informed consent for case presentation and publication was obtained from the patient and her parents.

Case Presentation

A twelve-year-old, E.N., applied to the dermatology clinic due to hair loss and alopecia areas on the scalp resulting from hair-pulling behavior during sleep, her examinations were performed, and she was referred to child and adolescent psychiatry with a prediagnosis of sleep-isolated TTM. Hair-pulling behavior during sleep had been present for about 4 months and it was happening at night two or three times a week. E.N. stated that when he woke up in the morning, there were broken hairs on the bed, but she was not conscious of pulling out, she did not wake up before and after plucking, and she did not remember anything about this behavior. Hairs on other parts of the body were unaffected. There was no other complaint other than pulling out hair during sleep. There was no hair-pulling behavior while awake and history of trichophagia. During the night during sleep, the patient's complex behaviors, unusual movements, nightmares, vivid dreams, confusional arousal, somnambulism, bruxism, leaping, screaming with sudden awakening, or other sleep experiences suggestive of parasomnias were not reported and were not noticed by the family. There was no history of snoring and apnea attacks, daytime sleepiness, and naps during the day. Her family stated that they did not witness hair pulling in E.N.'s sleep, they only noticed it from the hair on the bed the next morning. The patient and her family did not report an increase in hair pulling during sleep when anxious or worried, and stressful life events that would initiate or exacerbate it at present or in the past. There was no change in behavior during the day, not recognizing her parents and loss of response to any stimulus, and/or convulsions. It was learned that she did not use nicotine, caffeine, or drugs to help her fall asleep in the evening or at night. It was reported that she went to bed around 22:00 in the evening, woke up at 07:00 in the morning, and did not feel tired when she woke up. There was no difficulty in initiating and maintaining sleep, daytime sleepiness, and fatigue.

No problems were reported in E.N.'s pregnancy and birth history, early neuromotor and social development, and there was no dysmorphic feature, microcephaly, or neurocutaneous markers. Her peer and social relations were non-problematic, and she did not exhibit any emotional or behavioral problems. She had no history of febrile and/or febrile seizure, sleep disorder, or chronic medical illness. The patient and her family did not give a direct history of sexual, emotional, or physical abuse, and there was no history of domestic violence and conflict. There was no family history of any medical disease or psychiatric disorder including trichotillomania.

No psychopathological finding was detected in her psychiatric examination. Her intelligence level was clinically normal. In her general appearance, there were hair loss and alopecia patches of approximately 5-6 cm in diameter in the frontoparietal region of the scalp. The patient did not wear a hat and cap or any other camouflage method to cover the alopecic areas. Her eyebrows and eyelashes were intact.

Based on the psychiatric examination and history, sleep-isolated TTM, parasomnia, a sleep-related dissociative disorder, and/or an organic neurological disease were considered in E.N. In the psychiatric examination performed according to the DSM-5 criteria, no psychiatric disorder was found other than sleep-isolated TTM. In the Sentence Completion Test, there were no features related to anxiety, depression, or trauma, and its content was mostly about pulling out

hair. The child Sleeping Habits Questionnaire score was 35. Revised Child Anxiety and Depression Scale–Child and Parent Form scores were not significant. A sleep video was requested from the family, but the family could not provide it. Although parasomnia was not considered in the foreground, sleep electroencephalography and, if necessary, polysomnography were planned. Conversion disorder was excluded because seizure-like events characterized by tremor-like movements in the body and other complaints suggestive of conversion disorder were not described in the patient. Again, dissociative disorders were excluded due to the absence of physical, sexual, or emotional abuse history and psychosocial, medical, and environmental stressors, the absence of dissociative amnesia and alter identities, and the absence of nocturnal dissociative features. It was thought that the behavior of pulling her hair during sleep might be due to an organic cause and she consulted to pediatric neurology. In the evaluation performed in pediatric neurology, hematological and biochemical examinations, neurological examination and brain imaging results were normal. However, bilateral central isolated sharp wave paroxysms (intermittent generalized epileptiform anomalies) were detected in sleep-wake electroencephalography. In the treatment, primarily focusing on epileptiform activity, the pediatric neurologist started levetirecetam 750 mg/day treatment and was followed up, and no psychotropic medication was given. At the end of the third week of clinical follow-up, it was observed that the hair-pulling behavior was completely eliminated without any significant side effects with regular use of levetiracetam and there was no hair-pulling behavior again. The polysomnographic recording was not performed because the patient had epilepsy and improved with treatment. The patient was last seen in the sixth month of treatment, and her remission and well-being continued without symptoms. At 6 month follow-up, the alopecia had completely resolved and her current medication was still levetiracetam 750 mg/day.

Discussion

The aim of this article was to present a case of secondary sleep-isolated TTM, which is seen as an ictal symptom of complex partial seizure, and its treatment, and to briefly review the literature. There was insufficient evidence to suggest that the presented case suffered from any of the DSM-5 psychiatric disorders. In addition, anxiety, depression, or identifiable stressors that may be associated with TTM were not present in the case. There was no dermatological problem. Therefore, we thought that there might be an organic disorder underlying hair pulling only at night, and we aimed to exclude neurological diseases.

Hair pulling in TTM typically occurs when the patient is awake and in a comfortable environment. However, in a 2007 study with dermatologists, 11% of doctors reported that they had encountered patients with trichotillomania, which only manifests during sleep and the results of this study show that sleep-isolated TTM is not uncommon [7]. Again, in the same study, it was found that in 20% of unexplained hair loss cases, hair pulling occurs only during sleep [7]. In addition, there are some case reports of sleep-isolated TTM, in which some TTM patients have reported that they unwittingly pulled their hair only while sleeping [6, 8-11]. Sleep-isolated TTM is defined as hair pulling that occurs only during sleep but is not remembered while awake [7, 9]. In case reports, sleep-isolated TTM has been more frequently described as a sleep-related disorder (in the form of parasomnia) or a sleep-related dissociative disorder (in the form of non-epileptic seizures) rather than a manifestation of a psychological condition, and a treatment strategy in this direction has been carried out. In a case report published in 2006, a case of a 24-year-old woman who plucked her eyebrows and eyelashes only during NREM sleep, did not remember this when she woke up, and did not pull any hair while awake was reported [9]. The sleep-isolated TTM in this case, who did not have any other unwanted sleep behaviors and anxiety, depression, eating disorders, or other psychiatric problems, responded well to imipramine, similar to several other NREM sleep parasomnias [9]. In another case report, an 11-year-old boy with sleep-isolated TTM and primary nocturnal enuresis was presented; imipramine 50 mg and eight-week behavioral approaches were shown to reduce nighttime hair pulling [8]. In the same case report, the authors suggested that hair pulling during sleeping could be categorized as parasomnia (rhythmic movement disorder while sleeping) [8]. Based on history and examination, however, hair-pulling behavior during sleep in our case is not a possible explanation for parasomnias. Again, in another case report, sleep-isolated TTM was reported in a patient hospitalized for alcohol detoxification, fluoxetine was administered for the condition, but no improvement was seen [11].

Also, there is evidence that sleep-isolated TTM is a sleep-related dissociative disorder and the patient unconsciously pulls hair while sleeping in this case [10]. In 2015, a case report showing that sleep-isolated TTM emerged as a dissociative phenomenon in a 41-year-old female patient was published [10]. Additionally, it was also noted in this case that there were a number of typical characteristics suggestive of dissociative disorders such as being a woman, having a history of childhood abuse, having psychiatric morbidity, and experiencing daytime dissociative symptoms [10]. In our case, a sleep-related dissociative disorder was excluded because there was no history of childhood maltreatment, no symptoms indicating dissociative disorders, and no psychiatric morbidity including major depression and conversion disorder, psychomotor agitation during sleep, and nocturnal dissociation symptoms.

In addition, it was also stressed that stereotypical paroxysmal events during sleep, like the hair-pulling behavior in our case, may be a sign of nocturnal seizures [14]. Therefore, it has been emphasized that comprehensive seizure follow-up and polysomnographic study are indicated in such cases with atypical history. Children who experience complex partial seizures experience ictal symptoms such as impaired consciousness, loss of consciousness, and carrying out repetitive movements, called automatisms. Examples of automatisms, which are repetitive motions include verbal signs (crying, laughing, moaning, repetitive speech, screaming), oral movements (chewing, lip smacking, swallowing), and manual activities (fumbling, head rolling, patting, picking at things, removing clothing, walking, coordinated movements, such as cycling of the legs or a swimming motion) [15, 16]. Some rare ictal behaviors in adults are anorexia nervosa, drinking water, sexual ictal symptoms, spitting, multiple personalities and related dissociative phenomenon, and schizophrenia-like psychosis [17-22]. In our case, hair pulling was the only ictal action that was described, there were no other automatism movements. To the best of our knowledge, only one case report has documented that TTM is an ictal manifestation of epilepsy, but in this case, the TTM behavior occurred while awake, not asleep, and carbamazepine was successful in treating the condition [12]. It has not been previously described that sleep-isolated TTM is seen as a manifestation of epilepsy.

There is no standardized treatment model for sleep-isolated TTM. Based on clinical features, benzodiazepines (such as clonazepam, diazepam), tricyclic antidepressants (such as imipramine, clomipramine), serotonin reuptake inhibitors (such as paroxetine), other antidepressants (such as trazodone) and melatonin can be used in treatment [8, 14].

The significant limitation of this case report is that a polysomnographic examination was not performed to rule out sleep breathing disorder, confusional arousal, other NREM parasomnias, and REM behavior disorder, and sleep videos were not provided. However, it is possible to claim that the rapid improvement in the clinical status of the case with levetiracetam treatment eliminates the need for polysomnography.

In conclusion, this case report demonstrates that, even while trichotillomania typically co-occurs with other psychiatric disorders in clinical practice, it can also appear as an epilepsy symptom. Especially in cases presenting with isolated TTM behavior during sleep, the existence of epilepsy should be suspected in the differential diagnosis in addition to parasomnias and a sleep-related dissociative illness, and it should be aware that the treatment to be applied to these patients may differ from the conventional trichotillomania treatment strategies. Thus, as in our case, a dramatic improvement in the child's behavior can be seen through early diagnosis and appropriate antiepileptic medication. Nevertheless, further research is required to better characterize this condition and identify an effective treatment.

Acknowledgments: The authors want to thank the patient and her family for collaborating in the study.

Informed Consent: Written informed consent for case presentation and publication was obtained from the patient and her parents, on the condition that the patient's anonymity must be preserved.

Conflict of Interest: The authors reported no conflict of interest related to this article.

Financial Disclosure: There are no financial, personal, or professional interests.

Author Contributions:

Beyza Bozok and Öykü Özdemir: Research idea, study design, patient care, literature review, drafting manuscript, final approval and accountability.

Ayla Uzun Çiçek and Halil Ural Aksoy: Case follow-up, supervision, manuscript preparation, final approval and accountability.

İlknur Ucuz and Elif Abanoz: Literature review, critical revision of the manuscript, final approval and accountability.

References

1. American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th ed. Arlington, VA: American Psychiatric Association, 2013.
2. Henkel ED, Jaquez SD, Diaz LZ. Pediatric trichotillomania: Review of management. Pediatr Dermatol. 2019;36(6):803-807. doi:10.1111/pde.13954
3. Comertoglu Arslan S, Uzun Cicek A, Ucuz I, Dogru H. Sociodemographic variables, clinical characteristics, and treatments in children with trichotillomania in terms of age and gender: a multicenter study. Nord J Psychiatry. 2022;1-10. doi:10.1080/08039488.2022.2052179.
4. Torales J, Ruiz Díaz N, Ventriglio A, Castaldelli-Maia JM, Barrios I, García O, Navarro R, González-Urbieta I, O'Higgins M, Jafferany M. Hair-pulling disorder (Trichotillomania): Etiopathogenesis, diagnosis and treatment in a nutshell. Dermatol Ther. 2021;34(1):e13466. doi:10.1111/dth.14366
5. Jafferany M, Mkhoyan R, Stamu-O'Brien C, Carniciu S. Nonpharmacological treatment approach in trichotillomania (hair-pulling disorder). Dermatol Ther. 2020;33(4):e13622. doi:10.1111/dth.13622
6. Tay YK, Levy ML, Metry DW. Trichotillomania in childhood: case series and review. Pediatrics. 2004;113(5):e494-e498. doi:10.1542/peds.113.5.e494
7. Murphy C, Redenius R, O'Neill E, Zallek S. Sleep-isolated trichotillomania: a survey of dermatologists. J Clin Sleep Med. 2007;3(7):719-721.

8. Görker I, Karasalihoğlu S, Öztürk, L. Pulling of hair while sleeping: A pediatric case. Medical Journal of Trakya University. 2010;27(2):215-217. doi: 10.5174/tutfd.2009.00841.2
9. Murphy C, Valerio T, Zallek SN. Trichotillomania: an NREM sleep parasomnia?. Neurology. 2006;66(8):1276. doi:10.1212/01.wnl.0000208516.19583.ad
10. Angulo-Franco M, Bush-Martínez A, Nenclares-Portocarrero A, Jiménez-Genchi A. Trichotillomania and non-epileptic seizures as sleep-related dissociative phenomena. J Clin Sleep Med. 2015;11(3):271-273. doi:10.5664/jcsm.4542
11. Goyal D, Surya S, Elder J, Mccall WV, Graham K. Sleep-isolated Trichotillomania (SITTM): A Case Report. Innov Clin Neurosci. 2016;13(1-2):40-42.
12. Managoli S, Vilhekar KY. Tricotillomania as an ictal manifestation of partial seizure in a 4-year-old girl. Indian J Pediatr. 2003;70(10):843. doi:10.1007/ BF02723814
13. Hansen A, Brask BH, Nielsen J, Rasmussen K, Sillesen I. A case report of an autistic girl with an extra bisatellited marker chromosome. J Autism Child Schizophr. 1977;7(3):263-267. doi:10.1007 /BF01539002
14. Singh S, Kaur H, Singh S, Khawaja I. Parasomnias: A Comprehensive Review. Cureus. 2018;10(12):e3807. Published 2018 Dec 31. doi:10.7759/cureus.3807
15. Wyllie E, Rothner AD, Lüders H. Partial seizures in children: clinical features, medical treatment, and surgical considerations. Pediatr Clin North Am. 1989;36(2):343-364.doi:10.1016/s0031-3955(16) 36653-6
16. Fine A, Wirrell EC. Seizures in Children. Pediatr Rev. 2020;41(7):321-347. doi:10.1542/pir.2019-0134
17. Signer SF, Benson DF. Three cases of anorexia nervosa associated with temporal lobe epilepsy. Am J Psychiatry. 1990;147(2):235-238. doi:10.117 6/ajp.147.2.235
18. Rémillard GM, Andermann F, Gloor P, Olivier A, Martin JB. Water-drinking as ictal behavior in complex partial seizures. Neurology. 1981;31(2):117-124. doi:10.1212/wnl.31.2.117
19. Ellison JM. Alterations of sexual behavior in temporal lobe epilepsy. Psychosomatics. 1982;23(5):499-509. doi:10.1016/S0033-3182(82) 73382-1
20. Lawley A, Khan HA, Hegde V. Multiple whistling seizures in temporal lobe epilepsy: A case report and review of the literature. Clin Neurol Neurosurg. 2016;151:61-64. doi:10.1016/j.clineuro.2016.10.011
21. Schenk L, Bear D. Multiple personality and related dissociative phenomena in patients with temporal lobe epilepsy. Am J Psychiatry. 1981;138(10):1311-1316. doi:10.1176/ajp.138.10.1311
22. Hecker A, Andermann F, Rodin EA. Spitting automatism in temporal lobe seizures with a brief review of ethological and phylogenetic aspects of spitting.Epilepsia.1972;13(6):767-772. doi:10.1111 /j.1528-1157.1972.tb05161.x